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Secondary care costs to the NHS for children born with unilateral cleft lip and palate, bilateral cleft lip and palate and cleft palate from birth to 10 years of age

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Abstract

Aims

The aim of this study was to undertake a micro-costing approach to determine the secondary care costs to the National Health Service (NHS) associated with the cleft care pathway of patients being treated from birth until 10 years of age for bilateral cleft lip and palate (BCLP), cleft palate only (CPO) and unilateral cleft lip and palate (UCLP). This was compared to NHS reference and tariff costs.

Methods

A care pathway was mapped out for a child born with BCLP, CPO and UCLP. 23 case notes of children born with orofacial clefting were identified. Retrospective data collection of episodes of care were recorded. Costs of care were calculated using local and national sources in order to compare against NHS reference and tariff costs.

Results

Mean costs for BCLP, CPO and UCLP were £17,004.09 (SD £7,361.83), costs £6,137.49 (SD £2,319.87) and £11,619.74 (SD £2,547.81) respectively. Costs in BCLP were the highest due to the increased surgical care required when compared with CPO and UCLP. CPO had the lowest mean cost due to the least surgical care required. Did not attend (DNA) rates were similar in each of the phenotypes ranging from 5% and 8% of total appointments missed.

Conclusion

Costs for provision of care for BCLP, CPO and UCLP from birth to 10 years of age are significant and can vary based on the surgical and outpatient care provided. Staff costs formed the largest proportion of costs identified.

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Without the help of each of the individuals mentioned, I would have not managed to carry out this project and I am very grateful for all they have done for me.

Authors declaration

I declare that the work in this dissertation was carried out in accordance with the requirements of the University's Regulations and Code of Practice for Taught Programmes and that it has not been submitted for any other academic award. Except where indicated by specific reference in the text, this work is my own work. Work done in collaboration with, or with the assistance of others, is indicated as such. I have identified all material in this dissertation which is not my own work through appropriate referencing and acknowledgement. Where I have quoted or otherwise incorporated material which is the work of others, I have included the source in the references. Any views expressed in the dissertation, other than referenced material, are those of the author.

SIGNED: Saleem Hasanally (Digitally signed) DATE: 21st July 2020

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Glossary of terms

ABG	Alveolar bone grafting
BCLP	Bilateral cleft lip and palate
BNF	British National Formulary
CLP	Cleft lip and palate
CP	Cleft palate
CPO	Cleft palate only
CRANE	Cleft Registry & Audit Network
CSA	Certified surgical assistant
CSAG	Clinical Standards Advisory Group
CSSD	Central Sterile Services Department
DNA	Did Not Attend
ENT	Ear Nose and Throat
GDPR	General Data Protection Regulation
GSTT1	Glutathione s-transferase
HRG	Health Resource Group
MDT	Multi-disciplinary team
MFF	Market Force Factors
NHS	National Health Service
NICE	National Institute for Health and Care Excellence
OMFS	Oral and Maxillofacial Surgery
PPE	Personal protective equipment
QALYs	Quality adjusted life years
SALT	Speech and language therapy
TTA	To take away medications

UCLP	Unilateral cleft lip and palate
UHBW	University Hospitals Bristol and Weston NHS Foundation Trust

1. Introduction

Orofacial clefting is the most common craniofacial congenital condition found in humans (Slator *et al*., 2009) and presents in two main forms: cleft lip (with or without palatal clefting), or cleft palate (without lip clefting). Cleft lip and palate (CLP) is more common and is seen in 1-2 per 1000 live births. Cleft palate (CP) or cleft palate only (CPO) is seen in 1 per 1500-2000 live births. These relatively common congenital conditions are frequently associated with one of more than 400 distinct syndromes. This association is seen in approximately 30% of CLP affected children and 50% of CP affected children (Lidral *et al.,* 2008).

The care for cleft affected children involves a multidisciplinary approach with numerous interventions, often over a 20-year time period. A team approach involving nurse specialists, speech and language therapists, orthodontists, paediatric dentists, audiologists, Ear Nose and Throat (ENT), Plastic and Oral and Maxillofacial Surgery (OMFS) is required (Slator *et al.,* 2009).

The protracted nature of the treatment in terms of time, travel and number of appointments can be challenging for the child and their family in relation to the commitment that is required. In addition to this, the costs to the NHS are not inconsiderable. To date, there is no comprehensive assessment of the estimated costs associated with treatment of orofacial cleft affected individuals. Health economics models provide information to aid decision-makers in allocating resources where there is a finite quantity available. This requires clinicians and healthcare policy makers to prioritise healthcare needs, determining which care can, or cannot, be met due to financial constraints (Morris *et al.,* 2012). It is therefore important to evaluate the cost of cleft care to the NHS to improve budget allocation. Determining the costs could potentially allow a protected monetary reserve to be allocated to facilitate better quality care, by ensuring that departments are appropriately remunerated for the provision of care to cleft affected individuals. An economic evaluation would allow the cost of treatment to be weighed up against the perceived benefits (Drummond, 2015). At present, this is not possible as the true cost of cleft care is unknown.

2. Literature Review

2.1 Orofacial Clefting

2.1.1 Embryology of clefts

Clefting of the lip and/ or palate occurs when there is incomplete development or union of the orofacial structures *in utero*. In order to understand how clefts arise, and the likely problems and treatments an affected individual will encounter, normal orofacial development is described.

The cranial aspect of the embryo in humans develops relatively early on, with differentiation of cells beginning at around 20 days to form the neural systems. The three germ layers that initially form are the ectoderm, mesoderm and the endoderm, and it is these cells that differentiate to form the different types of cells and components required for development. The ectoderm eventually forms neural crest cells, which go on to make contact with the pharyngeal endoderm and mesoderm by the fourth week *in utero*, producing the five facial prominences:

- The frontonasal process

- The paired maxillary processes
- The paired mandibular processes

Towards the end of the 5th week of development the palate begins to form and is complete by week 12. The primary palate forms as a result of the medial nasal prominences fusing with the maxillary processes. It contains the maxillary incisors as well as forming the anterior hard palate and alveolar arch anterior to the incisive foramen. Secondary palate formation begins during the 6th week of development. The paired maxillary processes grow vertically down alongside the tongue bilaterally. As the tongue drops inferiorly during week 7, the palatal shelves migrate and sit horizontally above the tongue. The secondary palate fuses initially anteriorly and progresses posteriorly to completion at the uvular.

Clefts can arise as a result of disruption at any of these stages of lip and palate formation, with the final presentation dependent on the location and timing. Cleft lip and or palate occurs as a result of failure of the union of the lateral or medial nasal process and, or the paired maxillary processes (Figure 1).

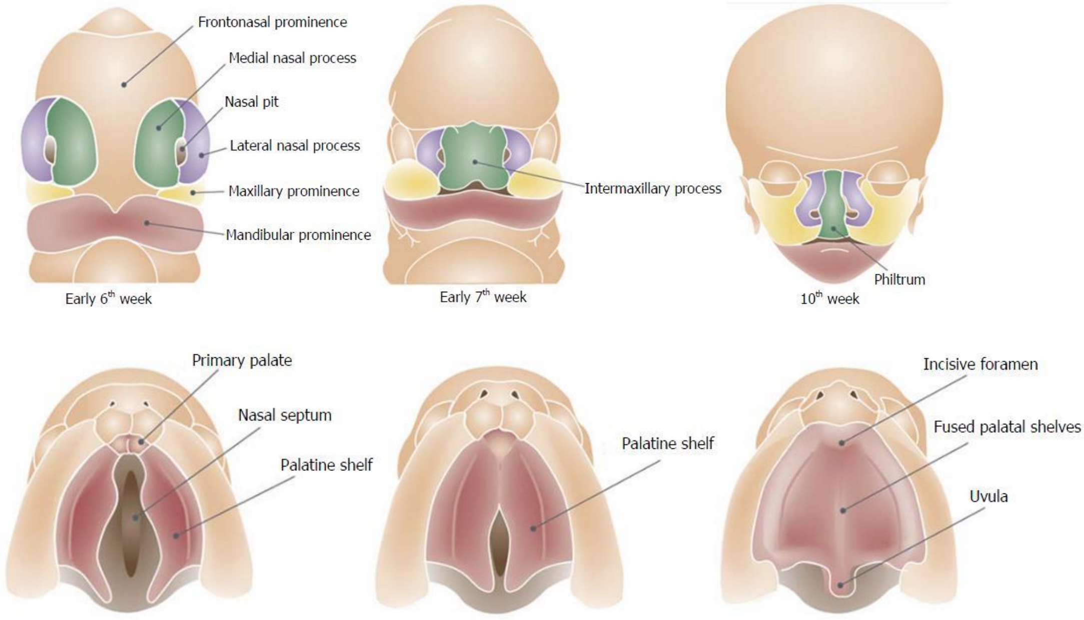

Figure 1. Development of the upper lip and palate in the 5th week of embryological development (Smarius et al., 2017)

2.1.2 Aetiology of clefts

Cleft lip and palate is a condition that can present as an isolated disruption during palatal or lip development, or may be part of a syndrome involving a number of additional phenotypic features. Most clefts are non-syndromic, with 70% of CLP and 50% of CP being non-syndromic and the remainder associated with one of over 400 different syndromes (Lidral *et al.*, 2008; Dixon *et al*., 2011). In the UK, cleft lip and palate affects around 1 in 700 live births, and worldwide approximately *"1.7 cases arise per 1000 liveborn babies, with ethnic and geographic variation"* (Mossey *et al.,* 2009).

Cleft lip and palate is a polygenic condition where genetic risk factors combine with environmental exposures (Dixon *et al*., 2011). Several genes have been linked to orofacial clefting (Table 1), including MSX1 (Jezewski *et al*., 2003), interferon regulatory factor 6

(Dixon *et al*., 2011), T-box transcription factor-22, poliovirus receptor-like-1, and P63 (Stanier and Moore 2004).

Polarising signals	Shh, Bmp2, Bmp4 and Bmp7, Wnt5a, Smad2-4
Growth factors and receptors	Egf, Egfr, Tgfa, Tgfβ1-3, Fgf1, Fgf8, Fgfr1, Fgfr2
Transcription factors	Ap2α, Dlx1-6, Gli2-3, Hoxa2, Irf6, Lhx8, Pax9, Pitx1, Prx1, Msx1, Tbx1, Tbx22
Cell adhesion molecules	Pvrl1, Connexin43, E-cadherin
Extracellular matrix	Col2A1, Col11A1 and Col11A2, Mmp2, Mmp3, Mmp9, Mmp13, Timp1-3, Fibronectin

Table 1. The "key genes required for craniofacial morphogenesis" linked to CLP (Stanier and Moore 2004)

Environmental factors have also been widely studied and several have been linked to the development of CLP, including:

1. Smoking, for which there is thought to be a weak but significant increased likelihood of developing CLP, with an increased relative risk of 1.3 to 1.5 (Kohli and Kohli, 2012). Specific genes in the infant have been shown to interact with maternal cigarette smoking, including the glutathione s-transferase (GSTT1), which increases the risk of CLP by almost 5 times (OR=4.9) (Rooij *et al*.,2001) and the MSX1, which increases the risk of non-syndromic clefts by almost seven times (Beaty *et al*., 2002).

2. Heavy maternal alcohol use has also been shown to increase the risk of developing a CLP by as much as 4.7 times (Munger *et al*., 1996). Low level alcohol consumption has not been shown to increase the risk of orofacial clefting (Natsume *et al*., 2000).

3. The relationship between maternal folic acid intake and CLP is not fully understood but the risk of developing a cleft may be tripled if vitamin supplements (mainly folic acid and cobalamins) are not taken during early pregnancy (Shaw *et al*., 2002). Although low level folic acid supplements are thought to have little or no preventative effect on the development of CLP (Ray *et al*., 2003), high daily doses (10mg/day) have been shown to reduce the risks by up to 65% (Kohli and Kohli, 2012).

4. Steroids are another environmental factor that have been studied due to their widespread use. A number of studies on mice have shown an increased incidence of CLP if glucocorticosteroids are given to the pregnant females (Melnick *et al*., 1981). Another study investigated maternal corticosteroid use in humans and found increased risks in humans for non-syndromic CLP (Carmichael *et al.,* 2007). Prednisone has been shown to increase the risk of CLP by as much as 3.4 times (Park-Wyllie *et al.*, 2000), which corroborates with evidence from animal studies (Kohli and Kohli, 2012).

5. Anticonvulsants have also been noted to increase the risk for congenital defects (Hernandez-Diaz *et al*., 2012) and include phenytoin/hydantoin, oxazolidinones, valproic acid and carbamazepine (Kohli and Kohli, 2012).

In summary it is clear there are both genetic and environmental influences on orofacial clefting, with interactions that are complex. The effects can be varied and are based on:

- The presence or absence of specific genes

- The type of environmental factors and concentration
- The stage of pregnancy when an environmental exposure has occurred

2.2 Classification of clefts

A number of different cleft classification systems are available. In 1922, Davis and Ritchie noted that articles relating to cleft lip and palate used different terms to describe the same conditions and felt the "*terminology (was) considerably confused*" (Davis and Ritchie, 1992). They decided that a three-group system would allow easier categorisation and used the lip, the palate and the alveolus as the segregations of the conditions. The alveolar process was to be the dividing line for this categorisation. The groups described were:

- Group I: Pre-alveolar process clefts (clefts that have lip involvement) – Further categorised as unilateral, bilateral, or median and either complete or incomplete.
- Group II: Post alveolar process cleft (palatal involvement) - Further categorised as hard or soft palate involvement
- Group III: Alveolar process cleft (alveolar process involvement) – Further categorised as unilateral, bilateral, or median and either complete or incomplete.

This system was predicated on a boundary "*the alveolar process forms the foundation for a surgical grouping*" (Davis and Ritchie, 1992, Allori *et al*., 2017). The system was criticised as being based on a surgical perspective, which is not a definitive way of treating the condition. Anatomy would provide a universally acceptable way of classifying the condition, regardless of what surgical procedures were carried out in different parts of the world.

Subsequently, Brophy in 1923 produced a classification involving 16 forms of cleft palate with or without lip involvement. This was seen to be impractical due to the complexity of the classification. In 1931, Veau simplified the classification system into 4 morphological forms:

- I – Soft palate clefts
- II – Soft and hard palate clefts, up to the incisive foramen
- III - Soft and hard palate clefts unilaterally extending through the alveolus
- IV - Soft and hard palate clefts bilaterally extending through the alveolus (Allori *et al*., 2017)

In 1942, Fogh-Andersen, a surgeon from Copenhagen published a new classification system in which he too adopted the incisive foramen as the division line for the different types of orofacial clefting. The groups were:

- Unilateral or bilateral clefting of the lip
- Cleft lip with cleft palate
- A cleft palate with no lip involvement
- Rare atypical clefts (for example midline clefting) (Allori *et al*., 2017)

The classification developed by Kernahan and Stark in 1958 classified orofacial clefting in line with the embryological formation of the cleft, rather than being centred around potential surgical correction. With the demarcation between the primary and secondary palates being the incisive foramen, a failure of formation of the “primary palate” (from the incisive foramen and anterior to it) would cause clefting of the lip and alveolus within the first 7 weeks of intra-uterine life. A failure of formation of the “secondary palate” (the hard and soft palates) in the subsequent 5 weeks would lead to clefting posterior to the incisive foramen.

Therefore, the new classification was as follows:

- Clefts anterior to the incisive foramen
- Clefts posterior to the incisive foramen
- Clefting anterior and posterior to the incisive foramen (Allori *et al*., 2017)

By 1976 Millard stated that no cleft system "*has been universally accepted because of language differences, inaccuracies, omissions and (a) lack of simplicity*" (Millard, 1976). This lack of a universal classification system not only meant that clinicians used broader descriptive terms of cleft lip, cleft palate and cleft lip and palate, but also meant that auditing and research was problematic due to the heterogeneous nature of the data (Allori *et al*., 2017).

Currently, most international cleft care teams use a pictographic notation, either the Kernahan striped-Y diagram, or written coded notations such as the LAHSHAL classification. The LAHSHAL classification was created in 1989 by Otto Kriens and was modified by the Royal College of Surgeons, England in 2005 to the LAHSAL classification (Figure 2). LAHSAL being a series of letters describing the 6 parts of the mouth that can be affected by clefting, namely: L – Right **L**ip, A – Right **A**lveolus, H – **H**ard palate, S – **S**oft palate, A – Left **A**lveolus, L – Left **L**ip.

If the cleft is complete, an upper-case letter is used (e.g. **S** – complete soft palate cleft). An incomplete cleft would be noted with a lower-case letter (e.g. **s** – incomplete soft palate cleft). No cleft is left blank. Therefore, a child with a unilateral cleft lip and palate (UCLP) would be classified as LAHS or HSAL codes, depending on whether the lip cleft was on the right or the left. This is the system used by the CRANE (Cleft Registry & Audit NEtwork) database and is used nationally in all UK cleft centres. The purpose of the CRANE Database, formed in 2000, was to collect information on children born with cleft lip and/or palate in England, Wales and Northern Ireland.

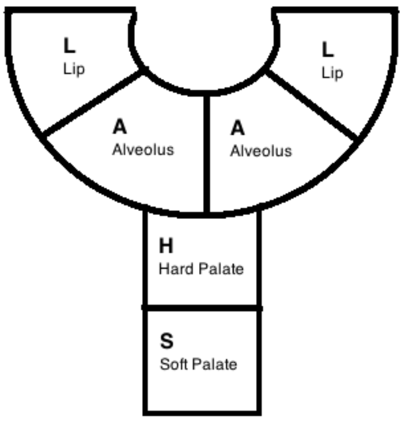

Figure 2. The LAHSAL classification used for the CRANE database. (image adapted from Hodgkinson et al., 2005).

2.3 Care pathways for each type of cleft

The care required for the cleft affected child differs to some extent according to the phenotype. This current project is focussed on unilateral CLP, bilateral CLP and CP only, and therefore only these care pathways will be described. The pathways may differ based on the region of the country and the choices of the local cleft team. However, they are broadly similar in terms of surgeries, timing of surgeries and the multidisciplinary teams involved.

2.3.1 Diagnosis and Birth

Early diagnosis is key and is one of the principal focusses of cleft care in the UK. Ultrasound is used routinely to detect the position of the placenta, the sex of the foetus and congenital abnormalities present in the foetus. Diagnosis by ultrasound sonography is a wellestablished method for detection of CLP and was first described in 1981 (Christ and Meininger, 1981). All women in the UK who are pregnant have a scan at approximately 20 weeks' gestation, which involves having a trans-abdominal ultrasound scan. Should any cleft abnormalities be found at this stage, the pregnant mother would be referred to the Cleft Lip and Palate Unit for counselling (Shaikh *et al*., 2001). Ultrasound is a non-invasive scan which has been found to cause no harm to the unborn child, whilst allowing the benefits of being able to prepare for the unborn child's needs. However, there are some problems associated with ultrasound scans. In particular, detection rates can vary significantly and may be affected by the experience of the sonographer. Detection rates for CLP range between 16 and 93 per cent (Smith *et al*., 2004) and an isolated CP is rarely diagnosed prenatally (Shaikh *et al*., 2001). Even if diagnosed, it is difficult to know if the cleft lip has alveolar or secondary palate involvement from an ultrasound alone (Smith *et al*., 2004).

There is evidence suggesting that prenatal diagnosis, with adequate support from the cleft team, can help the expectant parents to feel more psychologically prepared for the birth of their child, and better prepared and aware of the practical aspects of the child's care and the treatment pathways involved (Rey-Bellet and Hohlfeld, 2004). Previously, parents being told they are expecting a child with an orofacial cleft may have been more distressed, with rates of termination of pregnancy being affected by this knowledge (Chapman *et al*., 2003). More recently, a prospective cohort study has shown that more than half of parents expecting a child with oral clefting described it "*as a cosmetic disability*" (50.6%), or as "*just a little different*" (29.4%)." A minority (6.4%) considered termination of pregnancy, with none of the responders choosing to terminate pregnancy (Maarse *et al*., 2018).

When an unborn/born child with CLP or CP is diagnosed, a Cleft Nurse Specialist will contact the parents through either a telephone call or a visit to the hospital/family home within 24 hours. This is a good opportunity for the family to ask questions and understand what implications the condition may have for the child and the family. The initial referral process also starts at this stage. During the early days of life, feeding may be an issue for the child (Martin and Greatrex-White, 2013) and therefore feeding and oral care advice can be provided to the mother of the child (Beaumont 2012). Should there be complications, the multidisciplinary team can provide support as required, including referral to a dietitian or lactation team (Donovan 2012).

2.3.2 MDT (Multi-disciplinary team)

Following the recommendations of the Clinical Standards Advisory Group (CSAG) in 1998, to make cleft care a more centralised process (Sandy *et al*., 1998), 57 centres operating on orofacial cleft patients in the UK were reduced to just 11 centres, or managed clinical networks by 2011. The reason for this change was to improve outcomes for children affected with orofacial clefting. The current model is based on a hub and spoke, with the 11 centres or clinical networks (hubs) co-ordinating the treatment of patients from the smaller local hospitals (spokes). This ensures that surgeons who carry out the required procedures do so relatively routinely (at the hub), improving outcomes compared to operators with a limited caseload (Fitzsimons *et al.*, 2012). Outpatient appointments in many cases continue to be provided at the spoke units, in order to allow the children and their parents to travel easily to their appointments, minimising disruption to studies/work, and ensuring continuity of care with the same local team.

The cleft MDT comprises numerous professionals involved in the care of the CLP affected child. The team includes surgeons (maxillofacial, ENT, plastic surgery), dentists (paediatric dentists and orthodontists), speech and language therapists, a cleft nurse specialist and psychological support. Early exposure to the cleft team is advised, and generally occurs in the first 2 months of the baby's life. It is a good opportunity for the family of the baby to meet the different specialties and to allow patients to better understand what procedures are required and what forms of support are available for the baby and their family. There are specific cleft clinics run for audit purposes in order to ensure that the different aspects of the required treatment are being carried out in a timely manner, in order to give the best possible outcomes. These audit clinics are at five-year intervals, namely at aged 5, 10, 15 and 20 years.

2.3.3 Unilateral and Bilateral Cleft Lip and Palate

When a baby is born with a unilateral or bilateral CLP, the Cleft Team are contacted by the maternity ward and are referred the baby over the phone. The specialist cleft nurse will visit the baby on the ward and will discuss with the parents the nature of the cleft and the likely impacts. Feeding is assessed and often a different bottle/ teat is provided in order to aid this. Home visits are carried out after the baby has left the hospital in order to ensure that there are no concerns with feeding and that the parents are coping well. A referral is also made to the cleft clinic.

The first cleft clinic is within the first one to two months after birth and involves the baby being assessed by the cleft surgeons in order to plan the surgical repair of the cleft lip. The family are introduced to the different specialties including surgeons, orthodontists, paediatric dentists and audiologists. The family are also offered psychological support if required.

The first surgery, lip repair, is carried out at approximately three months of age and involves the baby staying in hospital with their parents for two to three days (one or two nights). Second stage surgery may be required (further lip repair in two stages in the case of a bilateral CLP and/ or scar revision of the initial lip repair) approximately three months later, which again would be two to three days (one or two nights) in the hospital along with their parents. Between the 9th and 12th months, the baby usually undergoes palate repair, also under general anaesthesia and this involves a two day (one night) stay in the hospital for the child and parents. Should any speech therapy or psychological treatment be required from the age of two years, treatment would commence as necessary. Orthodontic and audiology assessments begin at five years of age. Treatment with ENT/ audiology can involve grommets in children to improve hearing or customised hearing aid provision, whilst early orthodontic treatment will mainly involve simple orthodontic alignment of the teeth to a correct anterior crossbite. This can be done using an upper removable appliance when the upper permanent central incisors erupt at approximately six to seven years of age.

In the case of both UCLP and BCLP children, more complex orthodontic treatment is likely to be required to create space in preparation for canine eruption and associated alveolar bone grafting (ABG) at approximately 10 years of age. Grafting requires a hospital stay for the child as it is carried out under general anaesthesia (usually two days, one night in the hospital for the parent and child). Additional outpatient appointments are necessary immediately prior to and after surgery in order to assess the child prior to surgery and also to review the post-operative healing.

Orthodontic treatment is almost always required during the process of managing orofacial clefting. The role of the orthodontist in the multidisciplinary team is for record keeping, treatment planning and the provision of orthodontic therapy. This will usually be from soon after birth, through childhood to adolescence and beyond to adulthood (Cassi D *et al.,* 2017).

Facial surgery may be required for correction of a Class 3 skeletal pattern, when growth has just about ceased, at approximately 17-18 years old. This usually involves a course of presurgical and post-surgical orthodontic treatment in conjunction with either single or two jaw surgery under in patient general anaesthesia (usually a one or two night hospital stay). In some instances, an osteotomy may not be indicated, and orthodontic camouflage may be the appropriate treatment approach.

Care pathways have been mapped out for individuals with orofacial clefting (Figures 3 & 4). The following flow charts are from the South Wales South West Managed Clinical Network – Typical Patient Journey flowcharts (2016). They demonstrate care pathways for patients with a unilateral or a bilateral cleft lip and palate until the age of 10 years.

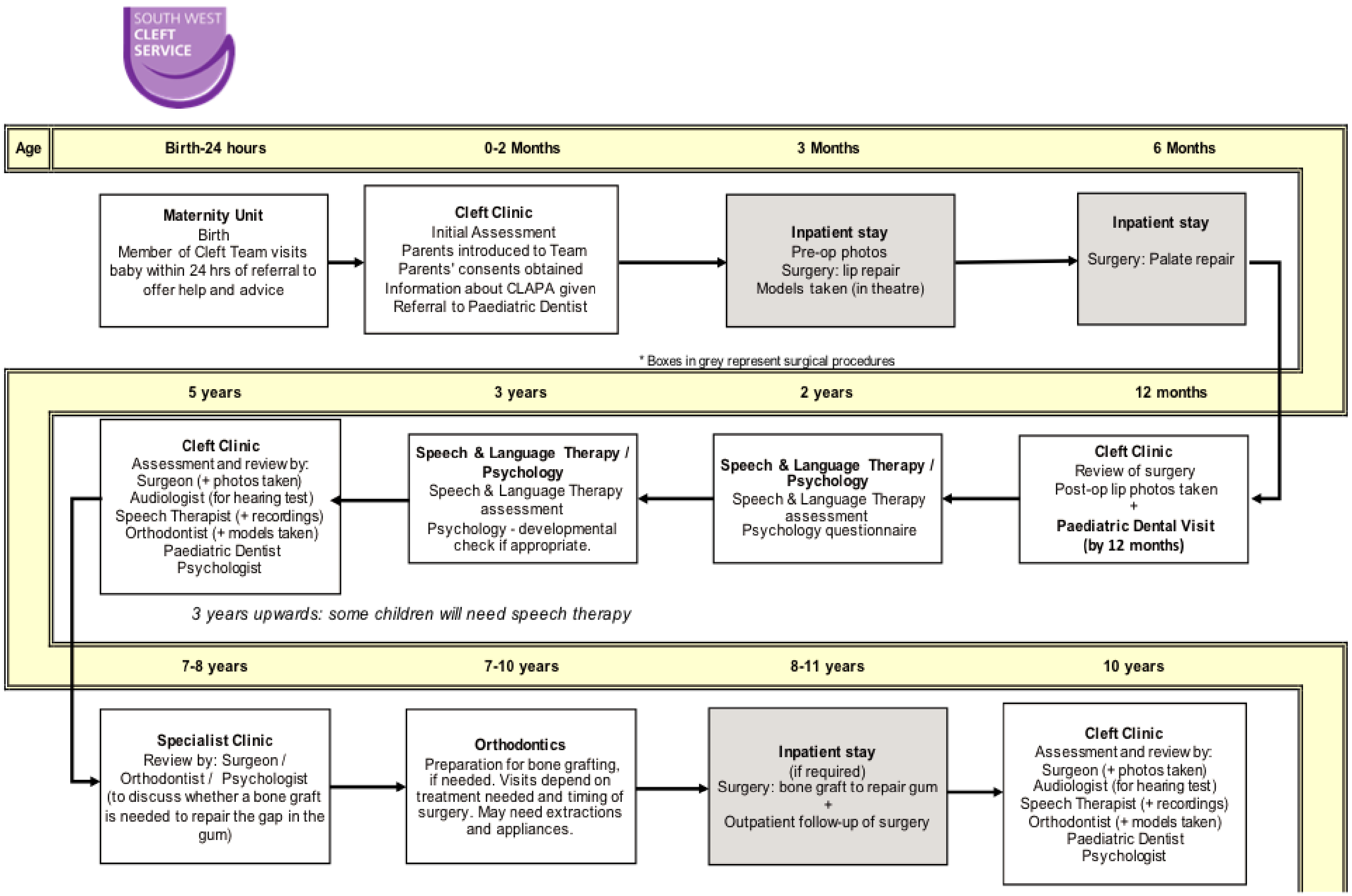

Figure 3. Typical patient journey for a child born with a unilateral cleft lip and palate (South Wales South West Managed Clinical Network – Typical Patient Journey flowcharts (2016))

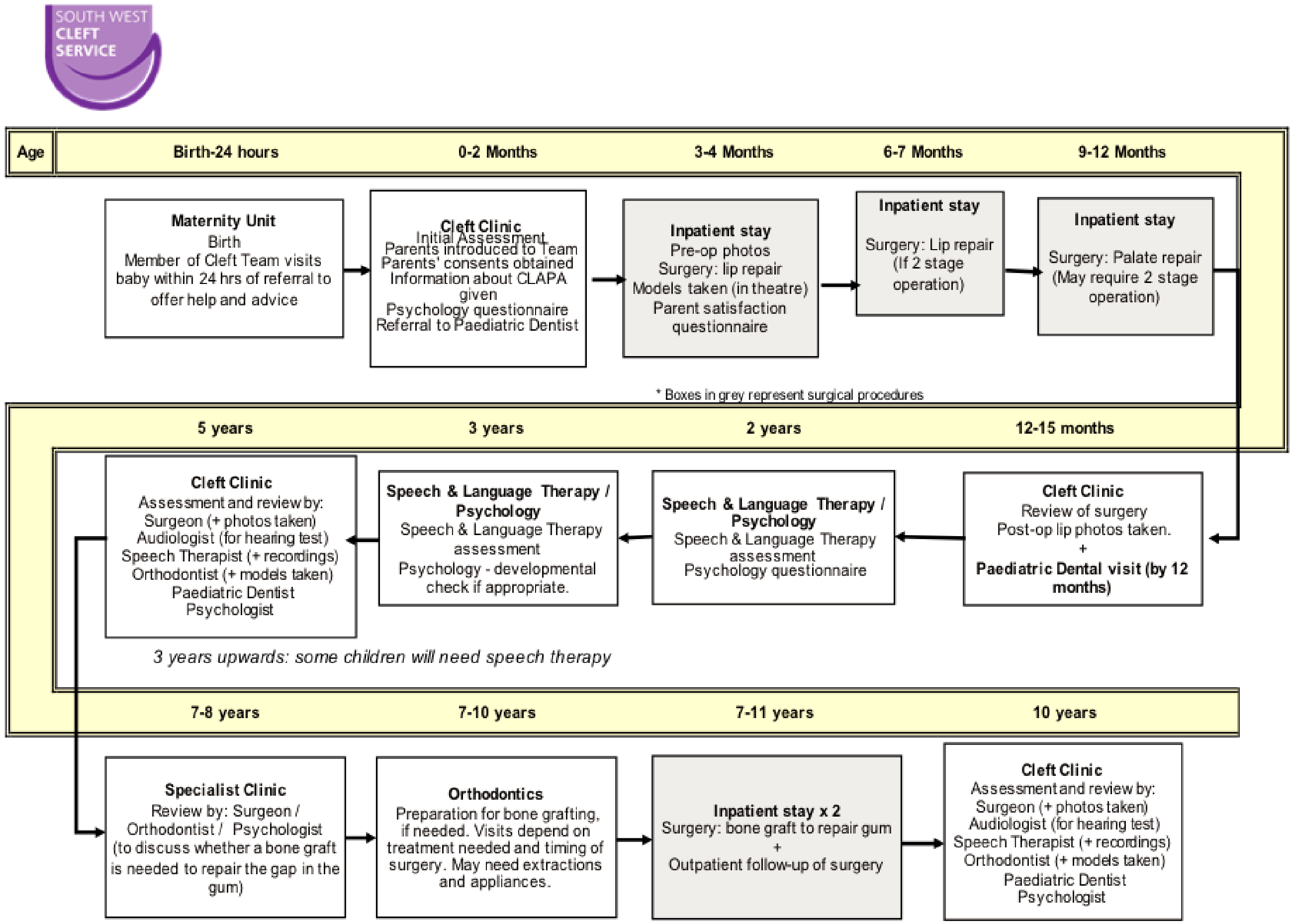

Figure 4. Typical patient journey for a child born with a bilateral cleft lip and palate (South Wales South West Managed Clinical Network – Typical Patient Journey flowcharts (2016))

2.3.4 Cleft Palate Only

An isolated cleft palate has a slightly less complex treatment pathway (Figure 5). As with cleft lip and palate, when the baby is born, a member of the Cleft Team initially meets the family in order to facilitate feeding and the referral process.

This is followed by an assessment in the cleft clinic, in which the baby is referred to the paediatric dentist and the family are offered psychological support if it is required. At 6 months of age, the baby has surgery to repair the palate, with a review being carried out 6 months following the surgery. This is different to treatment of cleft lip and palate, in which the lip repair is initially carried out at 6 months, followed by palate repair at between the 9th and 12th months.

The baby is periodically assessed on several cleft clinics, with psychological and speech therapy assessments beginning at two years of age as necessary. Orthodontic and audiology assessments begin at 5 years of age. Treatment, if required, would be carried out when planned to be most appropriate. This pathway contains fewer surgical interventions and therefore fewer hospital in-patient stays. Patients are seen by the paediatric dentist every 6 months to a year (unless otherwise seen in community) and cleft clinics at least once a year, including multidisciplinary team appointments at 5 and 10 years old at a minimum.

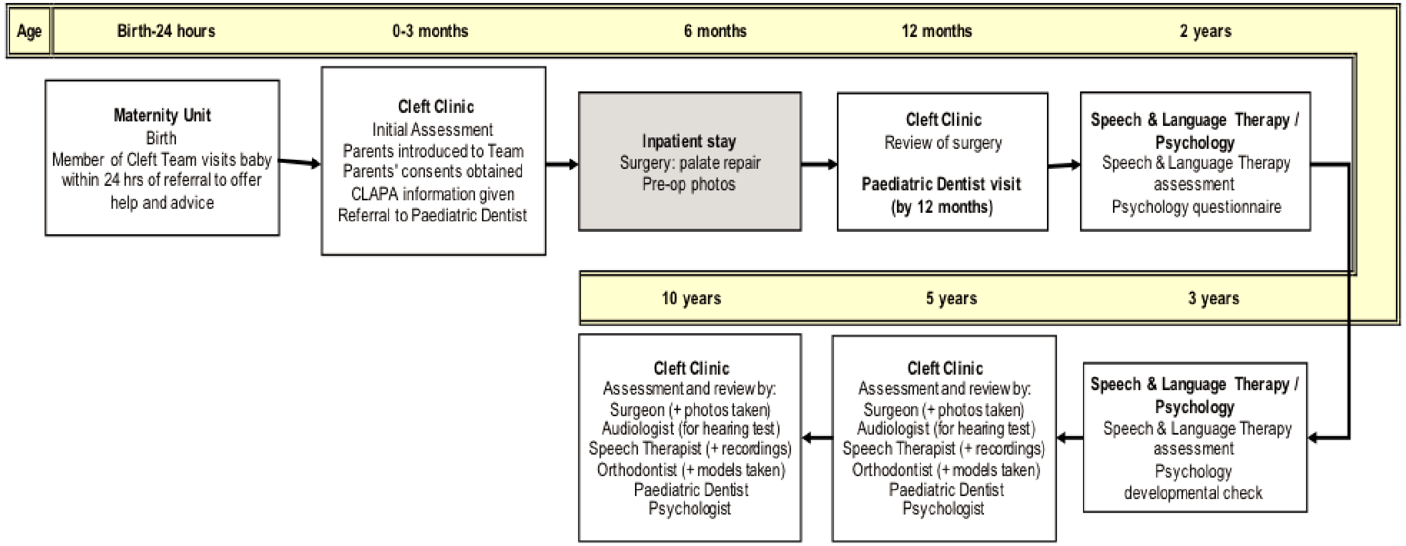

Figure 5. Typical patient journey for a child born with an isolated cleft palate (South Wales South West Managed Clinical Network – Typical Patient Journey flowcharts (2016))

2.3.5. Surgeries

The surgeries that CLP and CP affected individuals require are predominantly in the first year of life (including lip and palate repair) for primary surgery, followed by an alveolar bone graft at around 10 years if necessary. Primary repair may be carried out in two stages depending on various factors such as the size of the cleft, the health of the baby, and the success of the initial operation. This primary surgery may be carried out at different times around the world due to ongoing debates as to the best timing with respect to appearance and speech outcomes. Some care providers feel that surgery to repair the palate prior to nine months of age is most beneficial to speech outcomes. Others perform palate repair at 12 to 18 months of age to minimise any detrimental effects of surgery on growth (Sitzman *et al.,* 2017). In the UK, within the first year following birth, the initial surgeries are completed. This involves early repair of the lip, within the first three to four months, followed by the surgery to repair the palate ideally between six to nine months of age.

Historically, it has been said that "Millard's Rule of 10" should be followed with regards to carrying out surgery, *i.e.* infant's body weight of 10 pounds, haemoglobin count of 10g/dl, and older than 10 weeks. Much work has been done since showing that this should not be a rigid guideline, and that surgeries can be carried out earlier if required with good justification, especially with modern advances in paediatric anaesthesiology, which allow safer anaesthesia of infants (Ibadurahman and Sudjatmiko 2012).

2.4 Health Economics

Healthcare economics involves making decisions about allocation of resources where resources are finite, but demand is infinite (Morris 2012). These resources can be defined as time, money, materials and attention of the staff. All these resources could be used in other services or different industries. This results in a mismatch of what is available, compared with what is required, as the resources are limited. It is on this basic principle that we can better understand how the healthcare system works.

In the UK, healthcare is largely provided by the publicly funded National Health Service (NHS). The NHS is given a fixed budget from the UK government (money allocated from taxation), which is then assigned for a number of different healthcare applications. This includes staffing, materials, hospitals and clinics, research work, medicines and training of healthcare professionals. The NHS has commissioning and procurement teams and it also outsources contracts to non-NHS subcontractors for certain services. Currently the healthcare system is facing not only continually increasing costs for materials and services, but also an increased demand for its services. This is as a result of a growing population that is surviving far longer as a result of modern advances in medicine, with improved access to resources, and alongside the development of new drugs and treatments, which tend to be more costly.

Healthcare systems typically aim to maximise the efficient use of available resources, whilst providing the greatest benefits to the users of the healthcare services (the population). Health benefits can be measured in a number of ways, for example years of life saved, or quality of life changes. Money and time are spent on the healthcare system and it is important to analyse if this is providing good value for money.

The NHS, in the course of trying to provide a number of services, attempts to deliver each of these services in a cost-efficient manner, whilst taking into account quality (Buck, 2000). The latter can be measured in a number of ways. For example, markers of quality might include:

the number of surgeries that need re-visiting, length of hospital stays, infection rates, or quality of life improvements to name just a few.

As the NHS is a public-sector service, there is no direct monetary profit made from the achievement of better health in the UK. Although spending might decrease the need for further treatment and improve overall health, it can be difficult to identify as a direct financial benefit. As a result, healthcare systems have to use alternative measures, other than profit, to assess whether a service is being carried out efficiently, effectively and at a gain to the population, relative to spending on other potential services. Economic evaluation draws on the principle that if one service is provided, another service is unable to be provided due to limited resources and is defined as the "*comparative analysis of alternative courses of action in terms of their costs and consequences*" (Drummond *et al.* 2015). The opportunity costs in a healthcare system are defined as what "*could have been achieved had the money been spent on the next best alternative intervention or healthcare programme*" (Palmer and Raftery 1999).

Therefore, the distribution of available funds must be evaluated carefully in order to ensure resources are allocated appropriately and efficiently. If budget allocation and cost analyses are not carried out efficiently, there can be significant consequences. These can include waste, both in terms of *time* and *resource*, but also the *opportunity cost* of these resources: resources which may have otherwise been used to benefit other users of healthcare in the population.

Economic evaluation is a tool that helps optimise the distribution of resources spent in getting the maximum healthcare benefits in a financially constrained system. It is also important to appreciate that healthcare economics are based on a population and not on an individual basis. To maximise the health of the population, individual needs are not necessarily always met, as it the maximum health benefits to the population at large that are considered. Economic analysis is carried out by comparing resources used, against measured health outcomes, and compares alternative treatment methods and modalities, or different structures of providing care in terms of outcomes or costs. The main methods used to carry out economic analysis are (Kumar *et al*., 2006):

- Cost minimisation analysis
- Cost effectiveness analysis
- Cost utility analysis
- Cost benefit analysis

2.4.1 Cost minimisation analysis

Cost minimisation is generally used when costing is the main variable, with similar or unchanged outcomes (Robinson, 1993). This process involves assessing the costs of each of the individual interventions, allowing the most inexpensive intervention to be identified where there are similar outcomes (Kumar *et al*., 2006). Currently cost minimisation is discouraged, as it can be significantly biased where there is uncertainty of costs and large variations in the treatment costs. As a result, it can lead to an over or underestimation of costs. Instead, cost effectiveness analysis has been found to be more appropriate in avoiding biased estimations of uncertain costs (Dakin and Wordsworth, 2013).

2.4.2 Cost-effectiveness analysis

Cost-effectiveness analysis is useful in comparing interventions which may differ in terms of outcome, but have a standard unit of measure (Robinson, 1993). This process involves comparing the costs of interventions directly using common units, such as lives saved, days that pain killers were not required, or other common variables. This process relies heavily on assumptions that the patients may have all had similar experiences with the outcomes used, and only the one-dimension unit of comparison is the variable. As a result, it may be seen as a limited method. This is because it does not provide a true representation of the subjective experiences that individuals may have, and it does not allow comparison across disease areas, which is necessary for resource allocation (Kumar *et al*., 2006).

2.4.3 Cost-utility analysis

Cost-utility analysis is a subset of cost-effectiveness analysis and is a method of comparison of interventions which do not have a standard unit of measure of clinical outcomes (Robinson, 1993). Utility values are numbers representing "*parameters which influence a person's well–being* (Kumar *et al*., 2006)." Utility values are typically measured using the EQ-5D health-related quality of life tool (Clarke *et al.,* 2002). This tool evaluates quality of life and has population values assigned to each health state. The utilities are anchored at 0 and 1, where 1 is perfect health and 0 is death (Kumar *et al*., 2006). There are also negative values that are considered to be worse than death. This utility value can be used in a simple calculation to calculate the 'quality adjusted life years' (QALYs), which acts as a standardised unit of measure to be able to directly compare interventions (Drummond *et al*., 2015). QALYs combine both length of life and quality of life into a single number. Using QALYs to measure outcomes has been criticised due to certain treatments for non-life-threatening conditions being ranked higher in QALY than life threatening conditions.

The National Institute for Health and Care Excellence (NICE) has a role in helping the healthcare system deliver care to the best standards possible within the available resources.

NICE uses QALYs to assess if interventions are good value for money in terms of how much of an improvement a cost will make. However, it is not the sole way in which decisions are made, and should there be other grounds for an intervention to be used it may still be justified. QALYs are also a good way of measuring the benefits of treatments, whilst comparing different diseases and treatment modalities in a consistent way. Typically, NICE has mentioned an intervention which costs below £20000 per QALY to be cost-effective; if it was up to £30000, it would be cost effective if it could be justified (Ogden, 2017).

2.4.4 Cost-benefit analysis

The cost-benefit analysis is "*considered to be the most flexible method of economic evaluation*" (Robinson, 1993). This process is rarely used, particularly in the UK as the healthcare system is paid for by the state. Patients are not specifically "shopping" for healthcare and are thus unaware of the costs involved at times. As a result, "willingness-to-pay" studies, which can help cost outcomes, can be both difficult to conduct and inconsistent.

The process is carried out by calculating and placing values in monetary terms on the inputs and outputs of the system. In a healthcare system, the treatment costs are the inputs and the consequences are the outputs. This allows direct financial comparison of costs which are in the healthcare system, and costs which are outside of this system, such as environmental and education costs.

2.4.5 Economic evaluation of CLP

Within the current literature, there are a limited number of papers investigating the costs of cleft care. The first of these studies carried out a survey across the 16 UK cleft centres that were functioning at that time, on the management of otitis media with effusion in cleft palate affected children. It reported that 90% of children with CP have otitis media with effusion histories. The treatment cost per child (from cost year 2010-2011) varied between £593 (with no active treatment being carried out) and £2663 (insertion of ventilation tubes and hearing aid provision), depending on the choice of treatment (Bruce *et al*., 2015).

Another study by Mohiuddin *et al*. (2015), looked further into these costs by carrying out a cost-effectiveness analysis of treatment of otitis media using grommets in children with CP. This study adopted a decision tree model in order to assess surgical grommet insertion, with alternative options not involving surgery (*e.g.* hearing aids, or no active treatment). The study concluded that grommets were more cost-effective compared with hearing aids or antibiotic treatment methods. However, the study did not provide a measured cost of the treatment of otitis media in children with CP.

Another study observing CLP demographics, prevalence and complications in California over a 15-year period, with a yearly average case-load of 697 and a total number of correction surgeries over the 15-year period of 10,450, showed total median charges of $35,643 by the end of the study period in 2011 (Mahboubi *et al*., 2015). With revision surgeries being relatively common, this is a large expense. Although the results of this study are not likely to be directly comparable to the UK with its publicly funded NHS, it does illustrate that the provision of care for children with CLP may be associated with considerable costs.

By contrast, a study in Uganda of 343 cleft lip and cleft palate repairs, as a part of the training scheme for local surgeons by surgeons from overseas, the cost was just £27 per cleft repair. However, this cost did not take into account the expatriate staff salaries as the care was provided on a voluntary basis (Hodge and Hodges 2000). Therefore, the cost of care can vary greatly for different countries of the world and is likely due to many factors, including but not limited to differences in staff salaries, material costs, and the facilities where the care is provided in the different parts of the world.

Other studies in the USA have also attempted to analyse the costs of CLP associated surgeries. One study looked at 2,380 patients attending for cleft lip repair in 2010 and reported mean charges per patient for hospitalisation of $24,779 (Allareddy *et al*., 2014). The same authors had also carried out previous work into the factors associated with hospitalisation costs of cleft palate repair or revision, and reported mean hospital stays costs of $19,227. In this work, the authors describe a lifetime expenditure for orofacial clefts in the USA as being valued at $100,000, and describe not only might this be outdated, but that it can vary significantly with geographic location (Allareddy *et al*, 2012).

Another study attempted to carry out an analysis to assess the economic impact of cleft care in resource poor settings. A cost analysis, estimating how much impact was made by the surgeries carried out, assessed 1,142 reconstructive surgery cases over a 15-year period in Ecuador. Information was gathered from the logbooks supplied by "Hands Across the World," a charity for vulnerable children. Of these surgeries, cleft disorders constituted 277 cases, and 102 cases were individuals with primary cleft lip and/or palate. The study found that of the 102 cases of primary cleft repair "*between 396 – 1042 total disability-adjusted-life years were averted through surgery. This translates to an economic benefit between $4.7 million (human capital approach) and $27.5 million (value of a statistical life approach)*" (Hughes *et al*.,2012). However, this economic benefit approach does not provide a cost for the provision of care for individuals with CLP and is instead based on calculations of costs associated with disability-adjusted life years.

There is clearly limited evidence on the true costs of caring for children and adults affected by orofacial clefting and the costs will vary greatly across the world. To date no studies have effectively highlighted the costs of cleft care in the UK, nor indeed the world. The NHS provides care for patients with orofacial clefting, and it is important to know what this tertiary level of care provision costs the healthcare system, in order to make provisions for this service during budget allocation. It would be beneficial to assess the costs in order to predict what future costs may be.

3. Aims and Objectives

3.1 Aims of this study

The aim of this study was to undertake a micro-costing approach to determine the secondary care costs to the National Health Service (NHS) associated with the cleft care pathway of patients being treated from birth until 10 years of age for UCLP, BCLP and CPO. This was compared to NHS reference and tariff costs.

3.2 Objectives of this study

The objectives of this study were:

1. To identify the patient records at the Bristol Cleft Unit using the local database of children with cleft treated post CSAG from birth to 10 years of age.
2. Within this retrospective cohort of 30, obtain the records of 10 individuals with complete and incomplete unilateral cleft lip and palate (UCLP), 10 with complete and incomplete bilateral cleft lip and palate (BCLP) and 10 with cleft palate only (CPO).
3. To interrogate these records and use the data to identify the itemised costs of cleft care to determine the total cost of secondary cleft care to the NHS from birth to 10 years of age for each of the three cleft phenotypes
4. To compare costs identified with NHS tariff and reference costs

4. Materials and methods

Approval to carry out a cleft care service evaluation, with a view to reviewing the funding of the cleft service in the South West, was granted by the University Hospitals Bristol and Weston NHS Foundation Trust (UHBW) Caldicott Guardian on 19th April 2018.

The service evaluation was facilitated by the South West Cleft Team, and access to records was facilitated by the Clinical Director of the Cleft Team, Mr Scott Deacon. Previously mapped care pathways were used to assess the ideal pathway for an individual born with orofacial clefting. The pathways mapped by the South West Cleft Team for children born with UCLP, BCLP, and CPO from the ages of 0 to 10 years, with no associated syndromes, were used to carry out this cleft care service evaluation.

4.1 Materials required

- Laptop – Apple MacBook Air (password protected)
- Microsoft Excel, with a customised spreadsheet – Microsoft Corporation, One Microsoft Way, Redmond, Washington, United States, WA 98052-6399
- Secure storage device to keep data secure and compliant with General Data Protection Regulation (GDPR) – iStorage DatAshur PRO 256-bit USB Flash Drive

4.2 Inclusion

- Patients registered on the CRANE database with either UCLP, BCLP or CPO
- Patients who were over the age of 10 years, in order to be able to collect 10 years of retrospective data (birth to 10 years of age)
- Patients treated by the South West Cleft Team

4.3 Exclusion

- Patients who presented with a syndrome associated with the orofacial clefting

4.4 Care Pathway

Patients are seen within the cleft department at UHBW from birth. The cleft pathway (Figure 6) displays the journey of a patient who presents to the cleft team based on which one of the three phenotypes they present with.

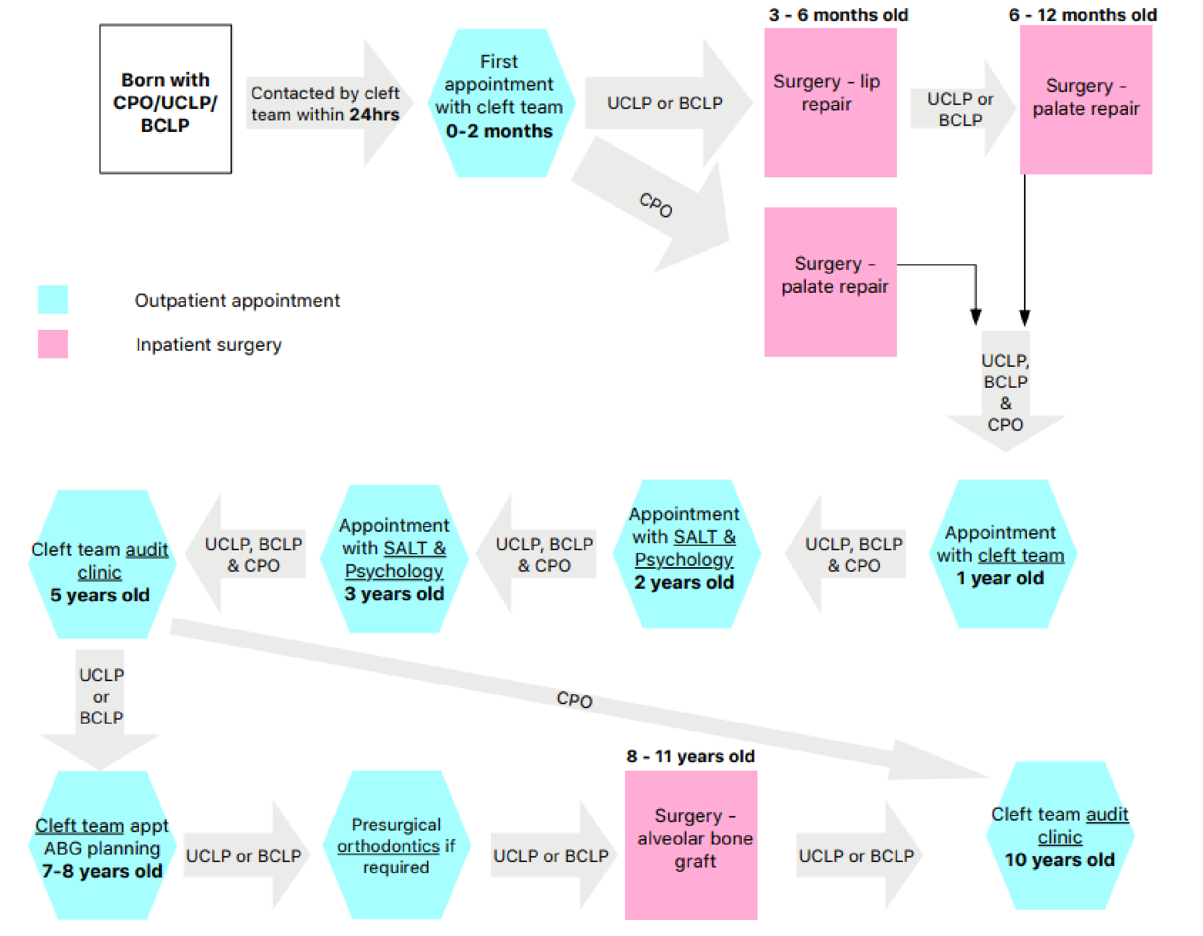

Figure 6. Simplified version of typical cleft care pathway of the three cleft phenotypes between ages of birth and 10 years old, modified from South Wales South West Managed Clinical Network – Typical Patient Journey flowcharts (2016)

4.5 Identification of patients

Although 10 individuals from each of the three phenotypes (30 in total) were to be studied only 23 could be identified using the agreed inclusion and exclusion criteria of the study applied when applied to the local source database (5 BCLP; 8 CPO; 10 UCLP). The clinical notes were requested from the central hospital records at UHBW with the help of the cleft department and obtained following study approval. The notes were initially securely stored in a locked room and were then transferred to the South West Cleft Team offices at the University of Bristol Dental Hospital for data collection.

4.6 Data collection

The hospital records for each of the patients involved in the study were interrogated in order to determine the number of hospital appointments they had for each of the different outpatient / inpatient specialties associated with cleft care. The following data were then collected and entered onto a Microsoft Excel spreadsheet and stored on an encrypted USB drive stored in a locked office:

a) Number of hospital appointments (and any relevant information):

- Cleft clinics (Single specialty)
- Joint cleft clinics
- Cleft nurse review (including inpatient, outpatient or telephone review)
- Speech and language therapy (SALT) (including school visits, home visits and telephone reviews)
- Psychology clinics

- Paediatric dentistry clinics
- Orthodontic clinic appointments

b) Surgeries involving cleft repair (including any medicines prescribed)

c) Duration of theatre time and recovery

d) Duration of inpatient visits due to cleft surgery

It became evident from letters and correspondence that much of appointment data was missing from the hospital notes, namely the SALT and Ear Nose and Throat (ENT) data. Following a discussion with the SALT team, it transpired they filed their own notes in their own department(s). Similarly, the ENT data was stored in a different unit within UHBW, namely St Michael's Hospital, rather than in the main UHBW records department. The missing SALT and ENT records were therefore requested and both the paper and digital notes were assessed.

Treatment for most of the children was carried out in the Bristol Children's Hospital. However, in certain cases, some of the treatment was carried out in the spoke hospital and in two isolated cases, treatment was carried out at other hospitals outside of the South West region due to family relocation. Due to the relatively low number of clinical notes available, these isolated cases had data collected where available. The durations of surgeries or the documented inpatient stays had the costs of the particular hospital applied, assuming that the duration of hospital inpatient stays or the surgery times would otherwise have been similar in other departments.

Some of the community-based treatment could not be assessed. Dental treatment and audiology were examples of such treatments that were sometimes carried out in the community setting, and sometimes in the hospital.

When there were any queries relating to the general data or logistics, advice was sought from the Clinical Director of the Cleft Service. In the event of any issues with audiology, SALT or ENT data, the SALT team were happy to help and provide direction as to how the queries could be addressed.

Where information was missing; advice was taken from the Clinical Director of the cleft service and Dr Thorn (Health Economist), one of the academic supervisors for the study. It was decided that missing appointments would not be estimated. Surgical missing information was estimated using collected averages of similar data. Where data was missing from the notes this was recorded as such on the spreadsheet.

Where possible, for each activity the following was recorded in the spreadsheet:

- The date
- The time required
- Any consumables used
- Whether it was inpatient or outpatient
- Pharmaceuticals used
- Letters written
- Staff present and grade
- Duration of the activity

Using this information, the costs for each individual activity were then calculated.

4.7 Staff costs

Staff costs are published by the NHS and can be accessed in the document "Agenda for Change" (NHS Employers, 2019). Using this document, a table was constructed in Excel to calculate the pay scales of the members of staff involved in the care of orofacial clefting.

Staff were all on different points of the salary scales in line with other responsibilities or levels of experience that they may have, and it was deemed to be inappropriate to ask them directly where they were on the pay scales. As a result, salaries from NHS job advertisements and discussions with both line managers and the Clinical Director of the Cleft Service were used to estimate the best cost salaries for staff and appropriately band their salaries within the NHS staff banding.

The yearly salaries of members of staff involved in the care pathway was divided by the number of working days in a year, which is 221 (as a result of annual leave, study leave and weekends), followed by dividing this by the working hours in a day. Salaries were placed into bands, or Consultant pay scales, and were categorised to the levels of experience.

The national insurance and pension contributions were also added to the salaries, in order to correctly calculate the cost to the employer for the staffing. This was found to be 14.38% in line with the NHS Business Services Authority. The salary per minute was therefore multiplied by 1.14, and a cost per minute for the staff was calculated from the annual salaries. The per minute salary was used in order to calculate costs of each of the clinical episodes.

The boundaries of the pay scales were used to calculate the upper and lower bounds of the staff costs and a midpoint of the salaries was also calculated in order to give an estimated range of what the cost might be.

Because this was a retrospective study, with clinical notes which did not have the durations of outpatient visits, or clinic durations, average clinical times and not exact amounts of time spent per patient were used. Outpatient appointments were discussed with the booking staff to assess the length of each outpatient appointment. Exact appointment times were not available in the notes, only the date of the activity and the associated clinical coding. Expert opinion was sought from the Clinical Directorate of the Cleft Service in order to estimate the length of clinic appointment times. The investigator also attended a joint clinic and appointment durations were assessed. This corroborated with the estimated times and so the booking times were assumed to be correct estimates of how long was spent with each patient. Appropriate departments were also consulted to accurately estimate if booked times were a true reflection of time spent on each patient. This was a pragmatic and robust way of calculating clinical appointment costs retrospectively.

Inpatient durations were calculated from entries in the clinical notes and were also used to estimate the nursing costs. Surgical theatre times were determined in order to calculate staffing costs per minute of surgical time. Although the surgery inpatient dates were present, sometimes no operating theatre times were recorded. In this case, an average of the surgical times and consumables was used, which was found from the data on similar procedures.

4.8 Consumables

Consumable costs were calculated for both inpatient and outpatient appointments for each patient. Consumables included costs associated with sterilisation of multiple-use items such as surgical instrument kits and costs of single-use items such as personal protective equipment (PPE).

4.9 Outpatient appointments

Sterilisation costs for outpatient appointments were found by meeting with the management of the Central Sterile Services Department (CSSD). As outpatient appointments involved using a sterilisable tray of instruments, the sterilisation cost for a tray of instruments was used as an estimated given cost associated with specific clinical appointments. The appointments chosen to have this assigned cost (cleft clinic, joint cleft clinic, single specialty clinic (including ENT and Orthodontics), speech investigation, cleft nurse review and paediatric dentistry) were elucidated by discussing with the clinical teams attending these clinical appointments. The management of the CSSD estimated that the costs for sterilised outpatient trays were approximately £20.

The other clinics such as the cleft audit clinics rarely used sterilisable clinical trays and this was therefore not an assumed cost for these other appointments. Single-use items for outpatient appointments such as disposable gloves for the clinician, or a disposable plastic mirror were also not added due to their low cost *i.e.* less than 20 pence. Indeed, they may be included in the overheads associated with some clinical areas, and not including them as a separate cost reduces the risk of double counting.

4.10 Inpatient and surgical appointments

All of the procedures carried out required different levels and types of equipment and consumables. To establish the cost for consumables used in surgeries associated with orofacial clefting, I visited the paediatric surgical theatres at UHBW where the surgeries were carried out.

Lists of equipment required for each type of procedure were provided by the theatre staff and included both multiple-use instruments, which require sterilisation, and single-use items. A list of sterilisation costs was obtained from the Head of the CSSD at UHBW for each piece of equipment. Costs for single-use items were found by the Head of the CSSD using previous order invoices. A theatre session involving orofacial cleft repair surgery was attended by myself in order to validate these equipment lists, from which an appropriate assumption of the costs could be made. These lists for each surgical procedure were then used to appropriately cost each of the surgical procedures. Time was added for buffer time around the surgery during which surgical checklists are performed and consent is checked/patient is sent for. When the theatre is cleaned, notes are written at the same time. This buffer time was deemed to be 40 minutes (20 minutes either side of the surgical time noted) and was discussed with numerous operating surgeons in order to ensure it was a fair representation.

4.10.1 Pharmaceuticals

After interrogating the notes, it was found that at outpatient appointments, not only were medications rarely prescribed, but when they were prescribed, they were usually analgesics, which are relatively inexpensive. As a result, only medications associated with inpatient hospital stays (due to surgical procedures) were considered.

Inpatient medications can be split into anaesthetic medication costs, and medications taken home by the patient following the hospital stay, *i.e*. 'to take away' medications (TTA).

4.10.2 Anaesthetic medications

Anaesthetic medications were noted on drug charts present in the clinical notes and were similar for the various surgical procedures carried out. Using the medications found in the notes, and following discussions with anaesthetists both during the surgical procedure observed, and an anaesthetic consultant from UHBW, a list of average medications was compiled (fentanyl (500mg), propofol (200mg), atracurium (50mg), ondansetron (4mg), dexamethasone (6.6mg), co-amoxiclav (1.2g), IV paracetamol (1000mg), Hartmann's solution (1 litre), sevoflurane (per hour)), which was applied to all of the surgical procedures, per patient, in order to keep costs standardised. The list of medications was costed using the most up to date British National Formulary (BNF) at the time of writing, using costs per ampule, except for sevoflurane, which is costed per hour (then calculated per minute).

4.10.3 TTA medication

The discharge summaries and clinical notes were used to determine the medications which were prescribed on discharge along with discussions with the cleft team. The medications were analgesics, antibiotics and a mouthwash (paracetamol (32 tablets), ibuprofen (24 tablets), chlorhexidine (300 ml bottle), co-amoxiclav 500/125mg (21 tablets)). Each medication was costed using the BNF in the same way as anaesthetic medications.

4.11 Overheads

These were determined with the help of the Finance Department at UHBW and included: property maintenance, disposal of waste, utility charges, portering services, provision of uniforms, laundry costs, catering, rents and charges and capital charges.

Highly detailed cost lists were provided by the finance department which included costs of staffing. Some staffing costs were reviewed and removed in order to prevent double counting. These included consultant costs, as consultant time was specifically accounted for when costing appointments or surgery.

4.11.1 Outpatient appointment overheads

These were determined by multiplying the specific clinic time in minutes by the £1.21/minute outpatient overhead cost provided by the Finance Department.

4.11.2 Inpatient appointment overheads

These were split into ward stay costs and theatre costs. Following discussions with the Clinical Theatre Manager at UHBW, it was found that following all cleft surgery, patients are taken to the same ward if paediatric (Ward E602). The Finance Department provided an overhead cost of £286.34 as a per night cost for this particular ward. This cost was multiplied by the number of inpatient nights recorded from the notes.

Theatre overheads costs provided by the Finance Department were found to be £2.21 per minute of theatre time. This cost was multiplied by the time spent for the specific surgical procedure and the time spent in recovery for each patient.

4.12 Total costs

Total costs were found by adding outpatient and inpatient total costs. Statistical analysis was carried out to calculate mean costs and standard deviations from mean costs. Total derived costs were then compared with national costs.

5 Results

The data were analysed using Stata version 16 (Stata Corp, College Station, USA) statistics package and are presented as summary statistics of the costs using means, medians, maxima and minima as well as percentages.

The results are presented under two main categories:

1. The overall cost of care for each of the three phenotypes (BCLP, CPO, UCLP).
2. Costs of inpatient and surgical care compared with outpatient appointments. Clinic appointments where patients ‘Did Not Attend (DNA)’ have been included in total costs unless specified otherwise.

The results are presented from the median point of staff salaries unless otherwise specified. Costs have been calculated using the minimum, median and maximum salary points for each staff type involved in the Cleft care pathway.

Prenatal costs are included in the cleft budget and have not been included in this study. This would require further data collection and appropriate ethics approval for the parents of the individuals with the clefting. It was therefore deemed to have birth as the start point of where costs would be calculated from.

5.1 Overall cost of care for each of the three phenotypes (BCLP, CPO, UCLP)

There was a large spread in the BCLP (5 cases) total mean treatment cost, with the mean cost £17,004.09 (SD £7,361.83).

A similar wide range in the total cost of provision of care was seen in the BCLP cohort and was also seen in the cases of both the CPO (8 cases) and UCLP (10 cases) cohorts, with mean costs £6,137.49 (SD £2,319.87) and £11,619.74 (SD £2,547.81) respectively. However, as might be expected, the standard deviations for CPO and UCLP are significantly less than for BCLP. Proportionately, UCLP displays the lowest standard deviation from the mean cost. The larger cohort size for the UCLP group mitigates outlier results and gives a standard deviation closer to the mean.

Table 2 provides the mean overall cost of care for children born with each of the three phenotypes from birth until 10 years of age (up to and not including the 10-year audit clinic).

Phenotype	Mean overall cost of care	Range of costs
BCLP	£17,004.09	£7,666.93 to £26,524.37
CPO	£6,137.49	£3,263.42 to £9,393.06
UCLP	£11,619.74	£7,665.61 to £16,038.72

Table 2. Mean costs and range of costs for total cost of care of the three phenotypes per individual from birth to 10 years old (using median salary costs).

5.2 Surgical and inpatient costs compared with outpatient costs

The mean costs for the two categories were identified for each of the three phenotypes. Figure 7 clearly illustrates that the surgical costs formed a greater proportion of the total costs across all three groups. The surgical and inpatient costs for BCLP made up 69.4% of the total costs compared with UCLP, which were higher at 74.4%, even though the actual cost was greater for BCLP. This is explained by the higher total outpatient costs for BCLP compared with UCLP (Figure 7). CPO surgical and inpatient costs were significantly reduced at 54.9%.

Total outpatient costs were similar for both CPO (£2,863.43) and UCLP (£2,973.78) respectively showing that the surgical costs for UCLP are generally higher (as we would expect as UCLP requires more surgical intervention (also illustrated in the mapped care pathways for each phenotype Figure 6).

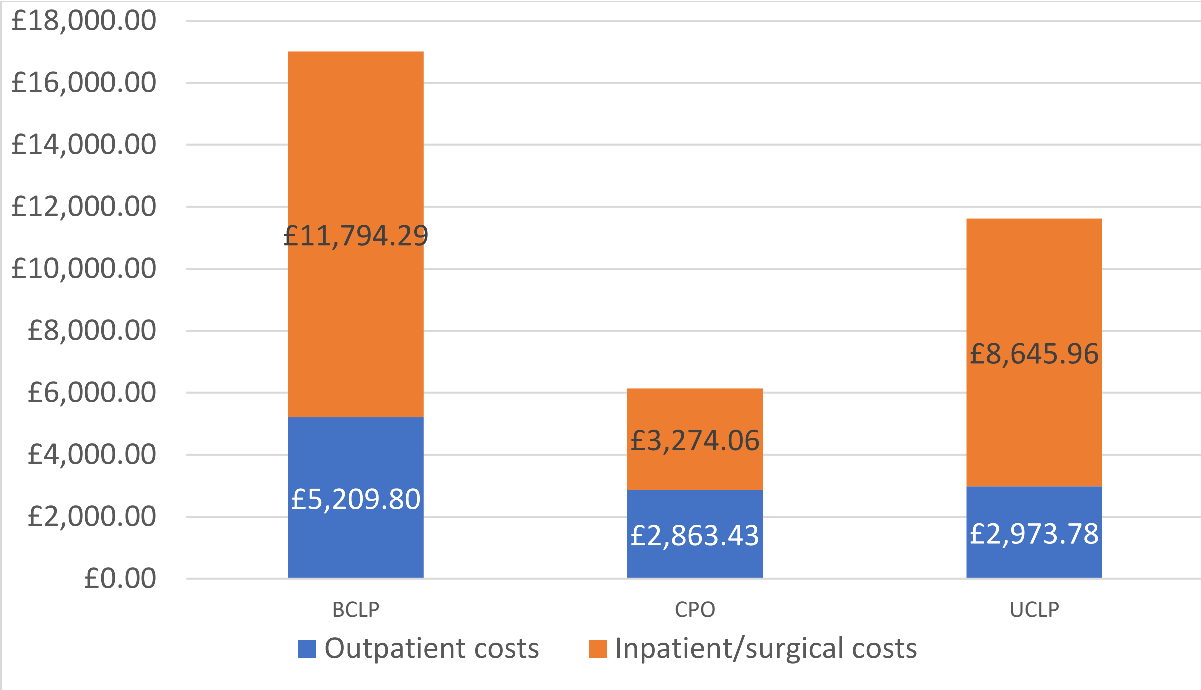

Figure 7. Mean costs (£) for outpatient and inpatient/surgical costs for each of the 3 phenotypes between birth and 10 years of age.

The overall mean costs for the provision of care from birth to 10 years of age for all three groups is shown in Figure 8. The variation between each group is specifically related to the salary points (minimum, median and maximum) of staff involved in the care of each of the phenotypes.

In the BCLP cohort, the lowest salary point for staff was a mean total of £16,237.51, whilst a maximum salary of staff equated to a £17,805.71 (£1,568.20 difference). For the CPO group these costs had a range from £5,894.54 to £6,382.91 (£488.37) and in UCLP these costs were £11,093.12 to £12,171.57 (£1,078.44). This reflects the reduced staff input in the management of UCLP and CPO when compared with BCLP.

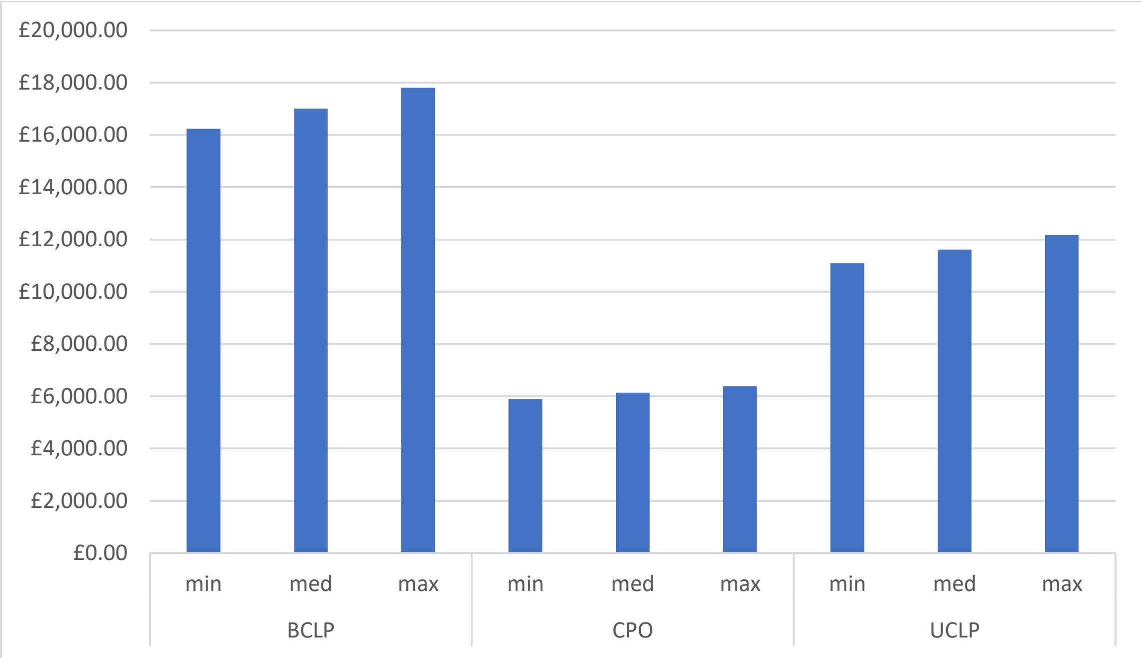

Figure 8. Overall mean cost of provision of care (£) from birth to 10 years of age (and not including the 10 years of age audit clinic) for minimum, median and maximum salary points.

Figure 9 illustrates the surgical cost for each patient with respect to the surgical interventions they underwent as part of their treatment, and for each of the three cleft phenotypes. There were some very obvious differences in the costs that can be attributed to phenotype. For example, alveolar bone graft (ABG) is carried out in BCLP and UCLP cohorts, but not CPO, although the costs for this are surprisingly similar for both BCLP and UCLP phenotypes. Bilateral lip repairs in BCLP cost more than unilateral lip repairs found in BCLP or UCLP. This would be expected due to complexity differences in the surgical procedures. In three of the five BCLP cases, bilateral lip repairs were not carried out. In these instances, two (separately timed) unilateral lip repairs were carried out in order to repair the bilateral lip clefting.

Palate re-repairs were found in all three phenotypes at similar costs. Grommets were least common in the BCLP group. Palate repair cancellations were found only in the CPO group and were charged at the bed cost for the night only. Palate repair was the most common surgery, as all patients would require this procedure to be carried out regardless of which phenotype they were, whilst lip repair would not be carried out in CPO.

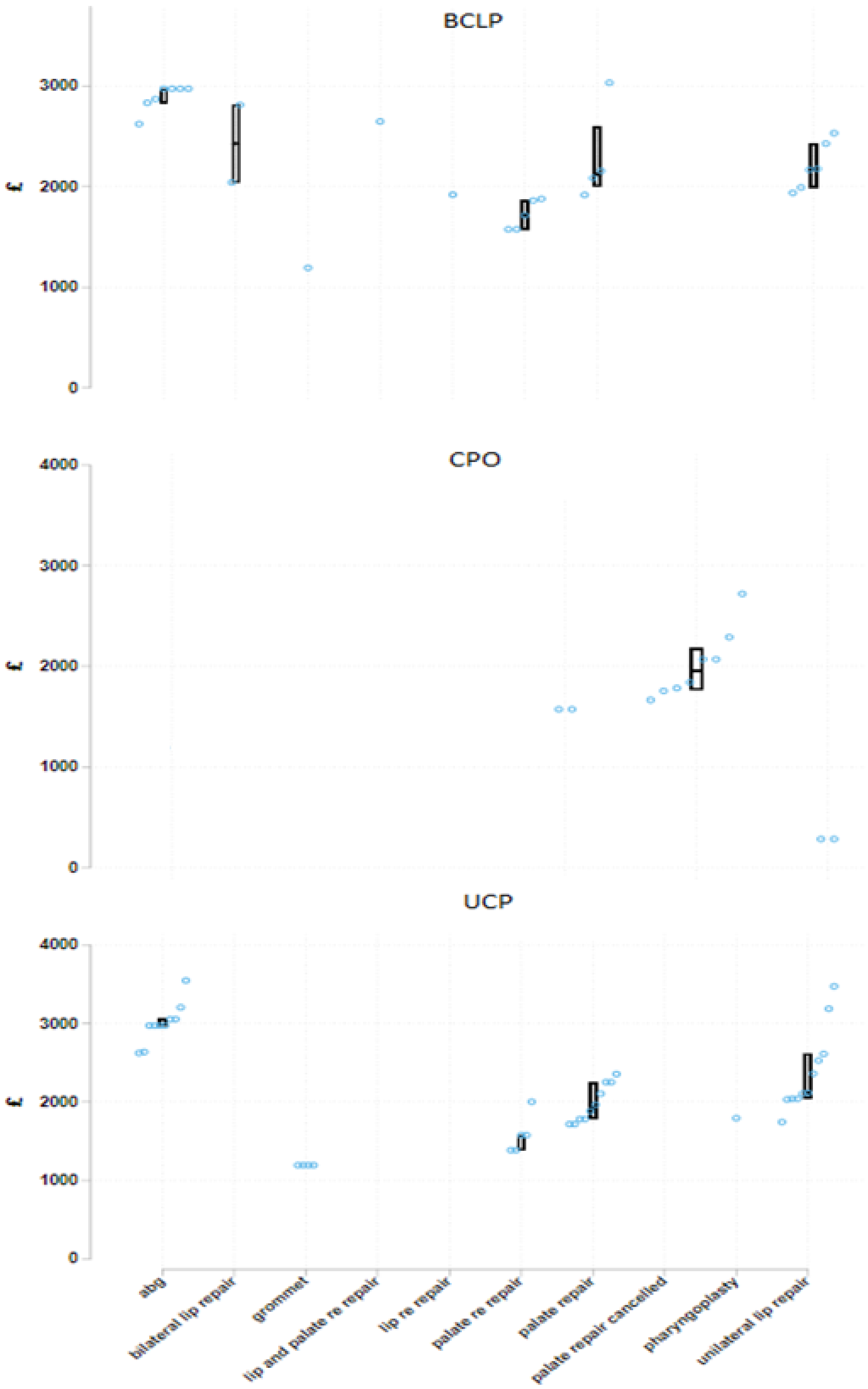

Figure 9. Surgery and inpatient costs plot (£) for each patient and for each surgery type within the three cohorts. BCLP costs are the highest total mean costs, with individuals needing a greater amount of surgical input. CPO costs are the lowest total mean costs as these individuals require the least total surgical input.

The average cost for each surgical intervention along with the attributed percent costs are shown in Table 3. It can be seen that staffing costs make up largest percentage of each of the surgical costs, ranging from 35 to 57% of the total cost of the procedures (£696.59 to £1439.86). Consumable costs were highest in the case of the ABG due to the materials required for augmenting a bone graft. All other surgical procedures had similar consumable costs. Anaesthetic consumables accounted for 1-3% of the costs of overall surgical procedures and were a fixed cost of £42.60. The bed cost of £286.34 (which was a fixed cost for 1 night) formed between 10 and 20% of the overall surgical cost. Overhead costs were calculated from the time taken to carry out each procedure and formed approximately 13% to 18% of the total mean cost of each of the procedures, ranging between £250.39 and £459.68.

Surgery	Number	Mean Cost	SD	Consumables cost for surgery		Anaesthetic consumables		Pharmaceutical TTA		Bed per night		Average overheads		Staff	
ABG	16	£2,824.30	£215.79	£1,099.59	38.93%	£42.60	1.51%	7.32	0.26%	£286.34	10.14%	£391.17	13.85%	£997.32	35.31%
Bilateral lip repair	2	£2,301.80	£532.79	£309.79	13.46%	£42.60	1.85%	7.32	0.32%	£286.34	12.44%	£459.68	19.97%	£1,196.11	51.96%
Grommet	8	£1,192.42	n/a	n/a	n/a	n/a	n/a	n/a		n/a	n/a	n/a	n/a	n/a	n/a
Lip and palate repair	1	£2,527.87	n/a	£309.79	12.25%	£42.60	1.68%	7.32	0.29%	£286.34	11.33%	£442.00	17.49%	£1,439.86	56.96%
Lip re repair	1	£1,837.05	n/a	£309.79	16.86%	£42.60	2.32%	7.32	0.40%	£286.34	15.59%	£331.50	18.05%	£859.54	46.79%
Palate re repair	12	£1,565.17	£184.18	£281.97	18.02%	£42.60	2.72%	7.32	0.47%	£286.34	18.29%	£250.39	16.00%	£696.59	44.51%
Palate repair	22	£1,953.37	£329.11	£281.97	14.44%	£42.60	2.18%	7.32	0.37%	£286.34	14.66%	£304.76	15.60%	£1,030.42	52.75%
Palate repair cancelled	2	£286.34	n/a	n/a	n/a	n/a	n/a	n/a		n/a	n/a	n/a	n/a	n/a	n/a
Pharyngoplasty	1	£1,713.88	n/a	£209.14	12.20%	£42.60	2.48%	7.32	0.43%	£286.34	16.71%	£309.40	18.05%	£859.12	50.13%
Unilateral lip repair	15	£2,204.85	£442.32	£309.79	14.05%	£42.60	1.93%	7.32	0.33%	£286.34	12.99%	£367.74	16.68%	£1,191.10	54.02%

Table 3. Average costs and percentage of total for each surgical procedure split into consumables, anaesthetic consumables, pharmaceutical drugs taken home by the patient after the procedure, cost of bed per night (assuming an average of 1-night stay), costs of average overheads and the staff costs.

5.3 Costs per outpatient appointment

Clinic cost breakdowns for each outpatient appointment variation are shown in a series of tables (Table 6 to 21) and show which members of staff were present in those clinics. The percentage of the total cost of each clinic is given for costs associated with:

- Staffing
- Overheads
- Consumables

Outpatient appointment durations were found by discussions with the booking teams and the cleft team. The booking times for each clinic type are shown in the following table (Table 4).

Clinical Appointment	Time in minutes
Cleft clinic	15
Joint cleft clinic	15
Single specialty clinic (including orthodontics and ENT)	15
Audit clinic	15
Speech investigation	15
Cleft nurse review	15
Cleft nurse phone call	10
Cleft nurse home visit	60
Paediatric dentistry	20
Audiology	15
SALT face to face	30
SALT telephone review	15
SALT school group session	60
SALT school or home	60
SALT and psychology	30
Photography	15
Psychology	30

Table 4. Time in minutes of each clinical appointment type

The staff present at each clinic are detailed in the following table (Table 5).

Clinical Appointment	**Staff**
Cleft clinic	1 consultant surgeon, 1 consultant orthodontist, 1 specialist nurse
Joint cleft clinic	1 consultant surgeon, 1 consultant orthodontist, 1 psychologist, 2 SALT
Single specialty clinic	1 consultant, 1 specialist nurse
Audit clinic	1 consultant surgeon, 1 consultant orthodontist, 1 consultant paediatric dentist, 1 specialist nurse, 1 psychologist, 2 SALT
Speech investigation	1 consultant surgeon, 1 consultant orthodontist, 2 SALT, 1 radiographer
Cleft nurse review	1 specialist nurse
Cleft nurse phone call	1 specialist nurse
Cleft nurse home visit	1 specialist nurse
Paediatric dentistry	1 consultant paediatric dentist, 1 specialist nurse
Audiology	1 audiologist, 1 nurse
SALT face to face	1 SALT
SALT telephone review	1 SALT
SALT school group session	1 SALT
SALT school or home	1 SALT
SALT and psychology	1 SALT
Photography	1 medical illustrator
Psychology	1 psychologist

Table 5. Clinicians present in each clinical appointment variety

Staffing costs associated with the ‘cleft clinic’ contributed to the majority of the costs associated with this outpatient appointment, with the percentage being 50.1% of the total clinic costs (Table 6). Overheads and consumables had comparable costs at 23.8% and 26.2% respectively.

Cleft Clinic			
Staff present	Cleft Consultant	Orthodontist	Specialist Nurse
Minutes	15	15	15
Staff cost – minimum salary point (£)	14.1	14.1	4.35
Staff cost – median salary point (£)	16.65	16.65	4.95
Staff cost – maximum salary point (£)	19.05	19.05	5.4
Overheads clinic cost (£)	18.15		
Outpatient tray sterilisation (£)	20		
Total clinic cost (med, min, max) (£)	**76.4 (70.7 – 81.65)**		
% Staff (median salary point)	50.1%		
% Overheads	23.8%		
% Consumables	26.2%		

Table 6. Total cost of 'cleft clinic' per outpatient appointment. Costs and percentage of clinic total cost shown for staffing, sterilisation of equipment and overheads.

The clinics associated with the highest mean costs were the ‘audit clinic’ and the ‘joint cleft clinic’ which had the most members of staff present. This is shown by the larger staff cost percentages of the joint clinic being 62.7% of the total clinic cost (Table 7), and the ‘audit clinic’ staff percentage being 82.7% of the total clinic cost (Table 8). The ‘audit clinic’ had 0% consumable costs as disposable mirrors and gloves were included in the overheads.

Joint Cleft Clinic						
Staff present	Cleft consultant	Orthodontist	Nurse	Psychologist	SALT	SALT
Minutes	15	15	15	15	15	15
Staff cost – minimum salary point (£)	14.1	14.1	4.35	11.25	5.55	5.55
Staff cost – median salary point (£)	16.65	16.65	4.95	13.5	6.15	6.15
Staff cost – maximum salary point (£)	19.05	19.05	5.4	15.75	6.75	6.75
Overheads clinic cost (£)	18.15					
Outpatient tray sterilisation (£)	20					
Total clinic cost (med, min, max) (£)	**102.20 (93.05–110.9)**					
% Staff (median salary point)	62.7%					
% Overheads	17.8%					
% Consumables	19.6%					

Table 7. Total cost of ‘joint cleft clinic’ per outpatient appointment. Staff present in the clinic have been costed, including two members of the SALT team. Costs and percentage of clinic total cost shown for staffing, sterilisation of equipment and overheads.

Audit Clinic								
Staff present	Cleft Consultant	Orthodontist	Paediatric Dentist Consultant	Psychologist	SALT	SALT	Audiology	Nurse
Minutes	15	15	15	15	15	15	15	15
Staff cost – minimum salary point (£)	14.1	14.1	14.1	11.25	5.55	5.55	5.55	4.35
Staff cost – median salary point (£)	16.65	16.65	16.65	13.5	6.15	6.15	6.15	4.95
Staff cost – maximum salary point (£)	19.05	19.05	19.05	15.75	6.75	6.75	6.75	5.4
Overheads clinic cost (£)	18.15							
Outpatient tray sterilisation (£)	20							
Total clinic cost (med, min, max) (£)	**105.00 (92.70 – 116.7)**							
% Staff (median salary point)	82.7%							
% Overheads	17.3%							
% Consumables	0							

Table 8. Total cost of 'audit clinic' per outpatient appointment. Staff present in the clinic have been costed, including two members of the SALT team. Costs and percentage of clinic total cost shown for staffing, sterilisation of equipment and overheads.

Speech investigation clinics involved five members of staff, of which one was a consultant with a significantly higher salary compared with the other members of staff present on the clinic. As a result, the staff costs were the highest proportion of the costs (51.2%) as shown in Table 9.

Speech Investigation (SPIN)					
Staff present	Cleft Consultant	Specialist Nurse	SALT	SALT	Radiographer
Minutes	15	15	15	15	15
Staff cost – minimum salary point (£)	14.1	4.35	5.55	5.55	5.55
Staff cost – median salary point (£)	16.65	4.95	6.15	6.15	6.15
Staff cost – maximum salary point (£)	19.05	5.4	6.75	6.75	6.75
Overheads clinic cost (£)	18.15				
Outpatient tray sterilisation (£)	20				
Total clinic cost (med, min, max) (£)	**78.2(73.25– 82.85)**				
% Staff (median salary point)	51.2%				
% Overheads	23.2%				
% Consumables	25.6%				

Table 9. Total cost of 'Speech investigation (SPIN)' per outpatient appointment. Staff present in the clinic have been costed, including two members of the SALT team. Costs and percentage of clinic total cost shown for staffing, sterilisation of equipment and overheads.

Cleft nurses play a pivotal role in the delivery of care to cleft affected individuals. This involves making initial contact with the parents at a prenatal stage and when the child is first born. Cleft nurses carry out face to face (Table 10), telephone reviews (Table 11), and home visits (Table 12). Face to face reviews costed more in consumables (46.4% of total clinic cost) than telephone reviews or home visits as clinical kits were used which required sterilisation. As a result, overheads carry a larger weighting in clinics with no consumable costs present and form the larger proportion of the costs (78.6%) in both the home visit and phone reviews. Home visits did not include travel costs because patients were living different distances away from the hub and this aspect would reduce the generalisability of the study.

Cleft Nurse Review	
Staff present	Specialist Nurse
Minutes	15
Staff cost – minimum salary point (£)	4.35
Staff cost – median salary point (£)	4.95
Staff cost – maximum salary point (£)	5.4
Overheads clinic cost (£)	18.15
Outpatient tray sterilisation (£)	20
Total clinic cost (med, min, max) (£)	**43.1 (42.5 – 43.55)**
% Staff (median salary point)	11.5%
% Overheads	42.1%
% Consumables	46.4%

Table 10. Total cost of 'Cleft nurse review' per outpatient appointment. Costs and percentage of clinic total cost shown for staffing, sterilisation of equipment and overheads.

Cleft Nurse Phone Review	
Staff present	Specialist Nurse
Minutes	10
Staff cost – minimum salary point (£)	2.9
Staff cost – median salary point (£)	3.3
Staff cost – maximum salary point (£)	3.6
Overheads clinic cost (£)	12.1
Total clinic cost (med, min, max) (£)	**15.40 (15 – 15.7)**
% Staff (median salary point)	21.4%
% Overheads	78.6%
% Consumables	0.0%

Table 11. Total cost of 'Cleft nurse phone review' per outpatient appointment. Costs and percentage of clinic total cost shown for staffing and overheads. Costs and percentage of clinic total cost shown for staffing and overheads.

Cleft Nurse Home Visit	
Staff present	Specialist Nurse
Minutes	60
Staff cost – minimum salary point (£)	17.4
Staff cost – median salary point (£)	19.8
Staff cost – maximum salary point (£)	21.6
Overheads clinic cost (£)	72.6
Total clinic cost (med, min, max) (£)	**92.40 (90 – 94.20)**
% Staff (median salary point)	21.4%
% Overheads	78.6%
% Consumables	0.0%

Table 12. Total cost of 'Cleft nurse home visit' per outpatient appointment. This does not include travel costs. Costs and percentage of clinic total cost shown for staffing and overheads.

Some patients were found to have numerous dental appointments with one BCLP case having 20 attended and 12 missed dental appointments, at a total cost of £2336 calculated using the median point of the salary scale. The median salary point was used because it was the midpoint between the highest and lowest salaries of staff of the same specialty as not all staff would be the most junior or the most senior. The largest cost component for this clinic (Table 13) was staffing at 39.5%, followed by overheads then consumable costs. Nine of the studied case notes had no dental appointments recorded. This reason for these data being zero appointments was that treatment may have been carried out in the community setting. The community data required was unobtainable for three reasons. First, the timeline for the project, second data protection guidance and third no ethical approval to obtain this information.

Paediatric Dentistry		
Staff present	Paediatric Dental Consultant	Dental Nurse
Minutes	20	20
Staff cost – minimum salary point (£)	18.8	5.8
Staff cost – median salary point (£)	22.2	6.6
Staff cost – maximum salary point (£)	25.4	7.2
Overheads clinic cost (£)	24.2	
Outpatient tray sterilisation (£)	20	
Total clinic cost (med, min, max) (£)	**73 (68.8 – 76.8)**	
% Staff (median salary point)	39.5%	
% Overheads	33.2%	
% Consumables	27.4%	

Table 13. Total cost of 'paediatric dentistry' per outpatient appointment. This does not include specific dental consumables e.g. restorative materials etc. Costs and percentage of clinic total cost shown for staffing, sterilisation of equipment and overheads.

Audiology costs were costed per clinic (Table 14). The majority of costs for Audiology clinics were consumables for sterilisation of equipment (40.6%). These clinics may involve laboratory fees other than the micro costed consumables costs, for hearing aid provision, hearing tests or other clinical activity. These additional costs were not measured. The notes were often difficult to interpret or were incomplete, and so a flat rate was costed for audiology clinics in order to prevent over costing. It is important to note that these costs can be substantial, with a study finding that the cost of a hearing test and provision of a hearing aid for one ear was £294, whilst provision of hearing aids for both ears totalled £388 (Campbell, 2015). In a study reviewing the hearing loss found in patients with cleft palate, it was noted that surgical (grommets) or conservative management (hearing aids) were both used to treat the hearing loss as a result of otitis media. However, hearing aids were the less frequently used method of treatment in 10.1% of cases (Gani *et al*., 2012). A number of the studied patients had grommet operations carried out and costs were assigned for this operation.

Audiology		
Staff present	Audiologist	Nurse
Minutes	15	15
Staff cost – minimum salary point (£)	5.55	4.35
Staff cost – median salary point (£)	6.15	4.95
Staff cost – maximum salary point (£)	6.75	5.4
Overheads clinic cost (£)	18.15	
Outpatient tray sterilisation (£)	20	
Total clinic cost (med, min, max) (£)	**49.25 (48.05 – 50.30)**	
% Staff (median salary point)	22.5%	
% Overheads	36.9%	
% Consumables	40.6%	

Table 14. Total cost of 'audiology' per outpatient appointment. This does not include specific treatment costs e.g. provision of hearing aids etc. Costs and percentage of clinic total cost shown for staffing and overheads.

The 'SALT review' appointment (Table 15), which is a face to face appointment was the most frequently attended overall. This had a (median salary point) clinic cost of £48.60. There were no overheads associated with this outpatient clinic and the overheads therefore contribute to 74.7% of the overall clinic cost. Staffing of this contributed to a smaller percentage of the total clinic cost at 25.3%, as there was only one member of staff present. This is the same breakdown of costs as a percentage of the telephone review (Table 16), group therapy (Table 17) or the school visits (Table 18). This was because the staff and overhead proportions were the same.

SALT face to face	
Staff present	SALT
Minutes	30
Staff cost - min	11.1
Staff cost - med	12.3
Staff cost - max	13.5
Overheads clinic cost (£)	36.3
Total clinic cost (med, min, max) (£)	**48.6 (47.40 – 49.80)**
% Staff (median salary point)	25.3%
% Overheads	74.7%
% Consumables	0.0%

Table 15. Total cost per 'SALT face to face' outpatient appointment. Costs and percentage of clinic total cost shown for staffing and overheads.

SALT telephone	
Staff present	SALT
Minutes	15
Staff cost - min	5.55
Staff cost - med	6.15
Staff cost - max	6.75
Overheads clinic cost (£)	18.15
Total clinic cost (med, min, max) (£)	**24.3 (23.70 – 24.90)**
% Staff (median salary point)	25.3%
% Overheads	74.7%
% Consumables	0.0%

Table 16. Total cost of 'SALT telephone review' appointments per episode. Costs and percentage of clinic total cost shown for staffing and overheads.

SALT group therapy. Four children in a group - priced per child	
Staff present	SALT
Minutes	60
Staff cost - min	22.2
Staff cost - med	24.6
Staff cost - max	27
Overheads clinic cost (£)	18.15
Total clinic cost (med, min, max) (£)	**24.3 (23.70 – 24.90)**
% Staff (median salary point)	25.3%
% Overheads	74.7%
% Consumables	0.0%

Table 17. Total cost of 'SALT group therapy' per appointment. As four children are in the group, this cost is for 1 child. Costs and percentage of clinic total cost shown for staffing and overheads.

SALT school visit or home	
Staff present	SALT
Minutes	60
Staff cost - min	22.2
Staff cost - med	24.6
Staff cost - max	27
Overheads clinic cost (£)	72.6
Total clinic cost (med, min, max) (£)	**97.2 (94.80 – 99.60)**
% Staff (median salary point)	25.3%
% Overheads	74.7%
% Consumables	0.0%

Table 18. Total cost of 'SALT school or home visit' per episode. This does not include travel costs. Costs and percentage of clinic total cost shown for staffing and overheads.

SALT and Psychology		
Staff present	SALT	Psychologist
Minutes	30	30
Staff cost – minimum salary point (£)	16.2	22.5
Staff cost – median salary point (£)	21.3	27
Staff cost – maximum salary point (£)	26.4	31.5
Overheads clinic cost (£)	36.3	
Total clinic cost (med, min, max) (£)	**84.60 (75 – 94.20)**	
% Staff (median salary point)	57.1%	
% Overheads	42.9%	
% Consumables	0.0%	

Table 19. Total cost per 'SALT & Psychology' joint clinic appointment. This does not include travel costs and is costed for a SALT consultant and a psychology consultant. Costs and percentage of clinic total cost shown for staffing and overheads.

Photography outpatient (and inpatient) activity was rarely recorded in the notes. Eight episodes of photographs were noted in four sets of case notes which were taken in audit clinics. If photographs were not seen in each of the clinical notes, they were not micro costed. Guidelines from the Institute of Medical Illustrators (Jones and Volcano, 2018) suggest that photographs for cleft individuals should be taken as a minimum at the first clinic attended and at the 5 years and 10 years audit clinics. These costs have not been added to the micro costing for each patient unless specifically noted as they were not evidenced. The cost of this appointment was predominantly the overheads cost at 74.7% of the clinic cost (Table 20).

Photography	
Staff present	Medical illustrator
Minutes	15
Staff cost - min	5.55
Staff cost - med	6.15
Staff cost - max	6.75
Overheads clinic cost (£)	18.15
Total clinic cost (med, min, max) (£)	**24.30 (23.70 – 24.90)**
% Staff (median salary point)	25.3%
% Overheads	74.7%
% Consumables	0.0%

Table 20. Total cost per ‘Photography’ appointment. Costs and percentage of clinic total cost shown for staffing and overheads. Costs and percentage of clinic total cost shown for staffing and overheads.

The costs of psychology input were found to be low across all three phenotypes. These clinics involved no consumable costs as staffing and overheads costs made up the costs involved with this clinic (Table 21). Psychology costs were accounted for in some outpatient multidisciplinary team clinics. These clinics included the joint cleft clinic, audit clinic and the joint psychology & SALT clinics. Patients or their parents may require psychological help during the provision of cleft care and this is often provided in the community setting. Community records would have required additional ethical approval being granted to access parents’ medical records and these have therefore not been costed. Only 12 single specialty psychology clinics (between 8 patients) were identified in case notes across all 23 patients studied.

Psychology	
Staff present	Psychologist
Minutes	30
Staff cost - min	22.5
Staff cost - med	27
Staff cost - max	31.5
Overheads clinic cost (£)	36.3
Total clinic cost (med, min, max) (£)	**63.30 (58.80 – 67.80)**
% Staff (median salary point)	42.7%
% Overheads	57.3%
% Consumables	0.0%

Table 21. Total cost of 'Psychology' appointments per outpatient appointment in the hospital setting. Costs and percentage of clinic total cost shown for staffing and overheads.

The costs for outpatients can be divided in accordance with each specialty outpatient clinic as shown in Figure 10. This is a total costs plot for each of the three phenotypes (BCLP, CPO and UCLP). The three groups had a varied number of patients in each of the groups and so the values are not comparable by cost value. The trends are however comparable. Figure 10 demonstrates that SALT was the highest outpatient expenditure for BCLP and CPO and the second highest expenditure in UCLP. SALT outpatient appointments include SALT home, group, face to face and telephone appointments. SALT & Psychology appointments have been categorised under the psychology grouping as psychology contributed to the bulk of this clinic staffing cost (Table 19). Cleft outpatient clinics were the second highest outpatient expenditure for BCLP and CPO respectively and the highest in UCLP. This included audit clinics, joint clinics and cleft clinics. It did not include cleft nurse reviews, which have been categorised separately as they are a substantial standalone cost. Orthodontic, psychology, photography and ENT clinic expenditure were noted as minimal in all groups.

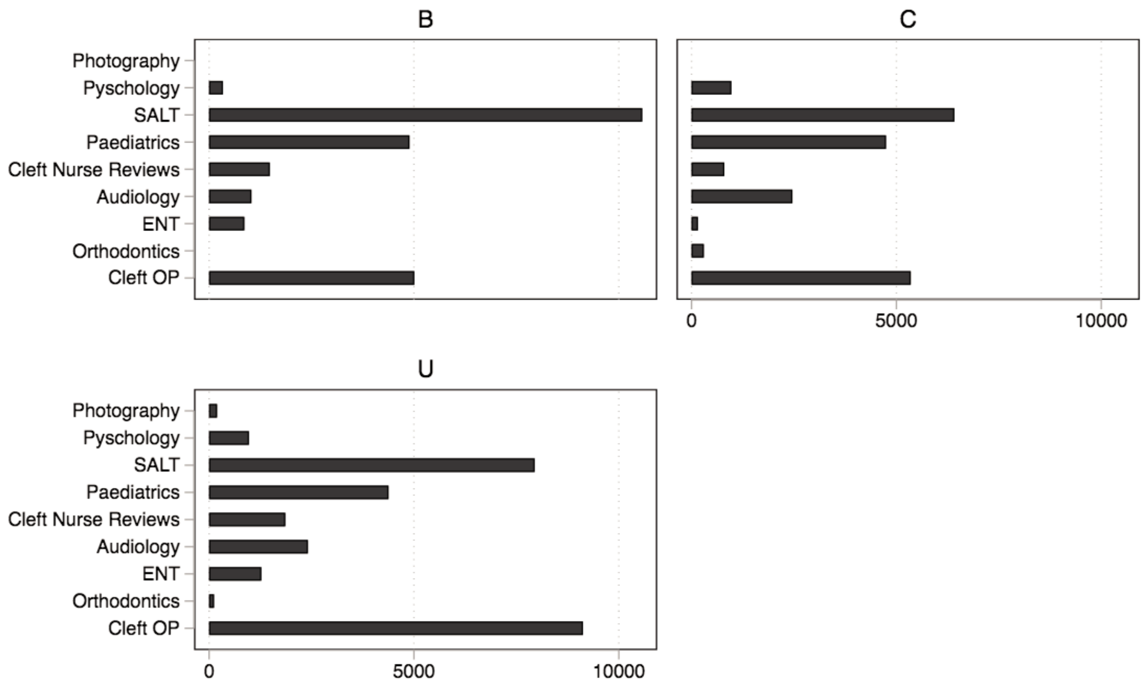

Figure 10. Total outpatient cost for each of the three phenotypes (B=BCLP, C=CPO, U=UCLP) split by cost for each specialty.

5.4 Missed appointments

Costs for missed appointments per phenotype are shown as a percentage on pie charts (Figures 11 - 13). The missed appointments as a percentage were similar across all groups ranging between 5 and 8%.

In the BCLP group (Figure 11), which comprised 5 patients, there were 25 missed appointments. Although this might suggest that on average each patient missed appointments, the range of ‘Did Not Attend’ (DNA) per patient was 0 to 14. DNA costs were assigned the same cost as a regular appointment.

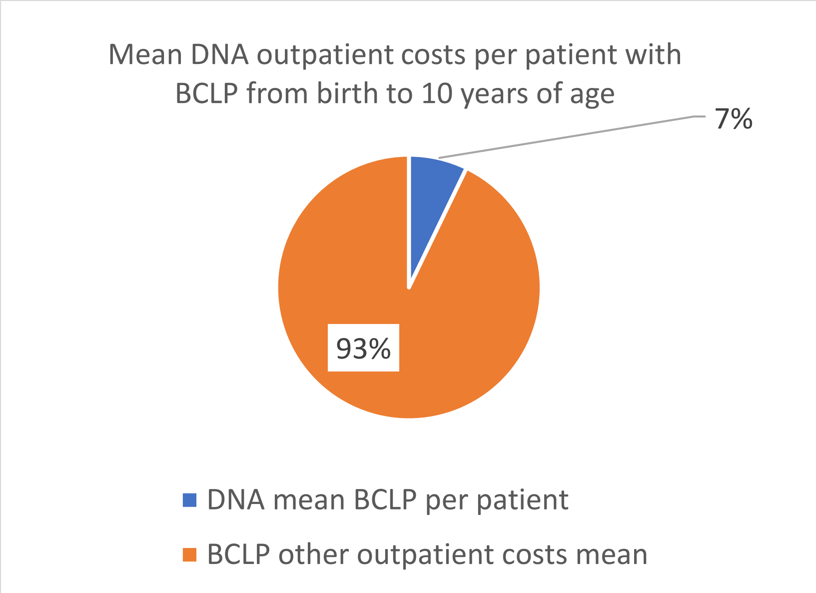

Figure 11. BCLP missed appointment (DNA) costs as a percent of total costs

For the CPO group (Figure 12), there were a total of 26 missed appointments, with an average of 3.25 DNA appointments per patient, ranging from 0-15 missed appointments.

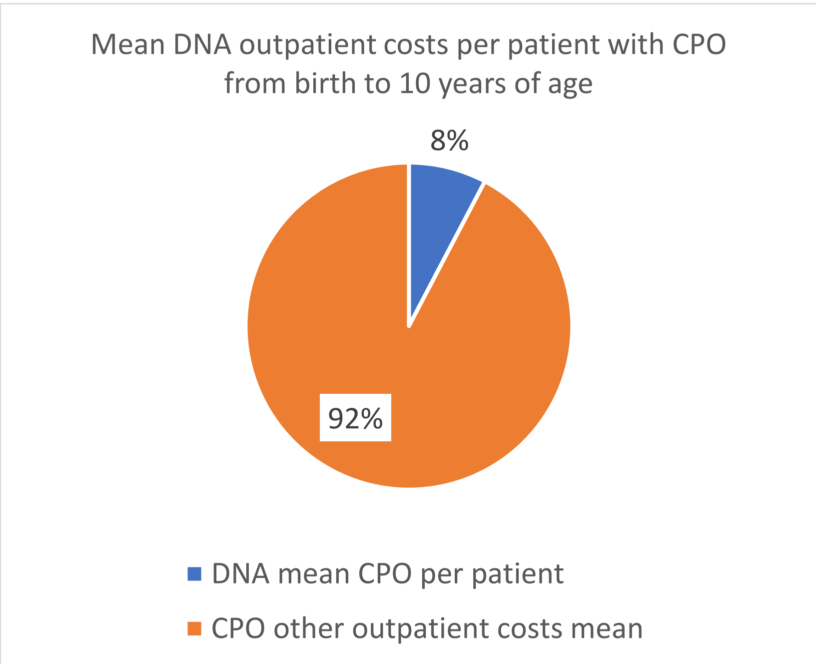

Figure 12. CPO missed appointment (DNA) costs as a percent of total costs

For the UCLP group (Figure 13), a total of 20 DNA appointments were found, with an average of two missed appointments per treated individual. The range of missed appointments was 0-12.

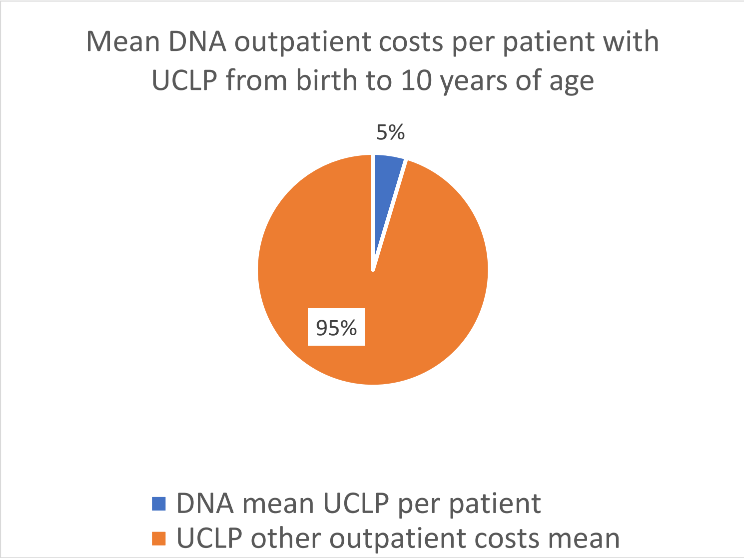

Figure 13. UCLP missed appointment (DNA) costs as a percent of total costs

5.5 Summary of results

The results show that mean costs for BCLP, CPO and UCLP were £17,004.09 (SD £7,361.83), costs £6,137.49 (SD £2,319.87) and £11,619.74 (SD £2,547.81) respectively. Costs in BCLP were the highest due to the increased surgical care required when compared with CPO and UCLP. CPO had the lowest mean cost due to the least surgical care required. DNA rates were similar in each of the phenotypes ranging from 5% and 8% of total appointments missed.

6 Discussion

6.1 Generalisability

This was a specific study of individuals born with cleft lip and/or palate between the ages of birth and 10 years old in cases with no evidence of associated syndromes. It is important to note that where patients have syndromes, costs may increase significantly due to differences in management. These differences, for example in cases with Pierre Robin sequence, can involve difficult airway management and may require the provision of a high dependency or a paediatric intensive care unit bed. This may require additional hospital review prior to discharge. Such cases may also take additional anaesthetist time to intubate and to manage the airway (Oxford handbook of clinical skills for children's and young people's nursing, 2012), and as a consequence would involve increased costs.

Some patients were seen in 'spoke' hospitals with consultants travelling from the 'hub' to pocket geographical areas. Travel costs have not been included in calculations due to difficulties in being able to accurately ascertain which staff were present in each clinic and individual variations of transport modality. Omission of this enables study generalisable to other departments by not factoring in travel costs at all.

One of the main advantages of this study is that the costs were calculated for the separate outpatient and inpatient/surgical aspects of cleft care for each phenotype. Thus, each dataset could be used to improve each aspect of the care pathway separately.

6.2 Participant cohorts

This study follows on from the work of Souster (2017) who carried out a similar micro costing of cleft care in UCLP between birth and 5 years of age. Although both his and the current study have found that costs were lower than NHS tariff, Souster only considered one phenotype, namely UCLP and recommended further work in this field. Recommendations for further work included extending the micro costing exercise to CPO and BCLP, ranging from birth to 20 years old. As a result, the current study, on three phenotypes from birth to 10 years of age, was planned and run with a simultaneous parallel micro costing study (Durman 2020 – not published at time of writing) of the same 3 phenotypes (UCLP, BCLP and CPO), but covering the ages 10 to 20 years of age on a different cohort of patients. The cut-off point of this study is up to but not including the '10-year audit clinic,' whilst Durman (2020) begins the study from the '10-year audit clinic.' The strength of running these two parallel studies on the same phenotypes but different cohorts, is that the data from both were from patients treated post the 1998 CSAG recommendations in the same centralised centre. This allowed the research data to be more comparable to other national data and covered the whole of the period an individual with a cleft will likely be in treatment, namely birth to 20 years of age.

In 1998, the CSAG report (Sandy et al., 1998) found that cleft outcomes were worse where operators were carrying out procedures infrequently. Recommendations from this report included forming a more centralised service. This centralised service involved a hub and spoke model in order to ensure that an adequate caseload was being seen by the fewer members of the team to aim for better outcomes due to each individual gaining more experience in the management of these types of cases. The changes from the CSAG report took a number of years to implement due to the significant structural changes to the system. Our decision to accept patients as close to 10 years old when recruited for this study ensured that the structural changes to the service had been implemented and therefore our data is comparable nationally.

This study was originally planned to include 30 patients, with 10 individuals of each of the three phenotype groups. Patients were selected if they met both the inclusion and exclusion criteria. Although only 23 sets of case notes were eventually assessed, due to these strict inclusion and exclusion criteria, it was felt that this is far outweighed by the advantage of having more contemporary research data that can be used to compare cleft data nationally and for future research if required. The numbers of studied cases for each of the phenotypes is shown in the table below (Table 22). All patients were aged 10 between 2015 and 2018.

Phenotype	Number of patients
BCLP	5
CPO	8
UCLP	10

Table 22. Notes obtained for the various phenotypes in this study.

6.3 Are the costs reasonable?

Prior to this study, there has been no research conducted to micro-cost the provision of cleft care within the UK from birth to 10 years old. There has been unpublished work which costed the pathway of UCLP from birth to five years of age by Souster (2017). This unpublished work was carried out in the same hospital studied here and found an average cost for treatment of UCLP to be £7076 per child for the first 5 years of care. For the first 5 years of treatment on the flowchart below (Figure 14), the mean micro costing of treatment can be seen to be £4,695.32, up to the 5-year audit clinic. However, this is a simplified care pathway and alongside this would be numerous other appointments with all of the other specialties individually, both in the hospital and community setting.

The current study has shown that the mean cost of provision of care for BCLP from birth to 10 years of age is £17,004.09 (SD £7,361.83), CPO is £6,137.49 (SD £2,319.87) and UCLP £11,619.74 (SD £2,547.81).

One of the reasons for large standard deviations of costs may be related to the variance in the amount of community care provided for some individuals, and that the micro costing study only accessed hospital records. A cleft affected child may require more or less input from the hospital service depending on the level of community care that is available, and they can access. This is an important point to note as some services, such as audiology and routine dental care, are often carried out in the community setting for the bulk of the population. Some patients may opt to have these aspects of care provided in the community for geographical convenience or for personal preference. However, if this is not possible, or the treatment or its management is slightly more complex, the patient may be assigned to have it provided within the hospital setting. The varying numbers of hospital outpatient appointments and the unknown number of outpatient appointment in the community setting were not costed and may therefore contribute to the wide range of costs. Only the hospital appointments were micro costed in this study. This might account for why some patients appeared to have many dental appointments, when others appeared to have very few or indeed none.

This wide range of costs makes it difficult to necessarily be able to assign a true mean value to the costs of BCLP, not only due to the reduced number of case notes assessed for this phenotype, which can mean outliers have a large effect, but also because the treatments were not costed within the community setting. A larger sample size would help to narrow the true mean costs, as would the ability to cost the community-based treatments.

The mean micro costed values have been applied to the care pathway to help visualise what the actual costs are for each of these aspects of care shown in Figure 14.

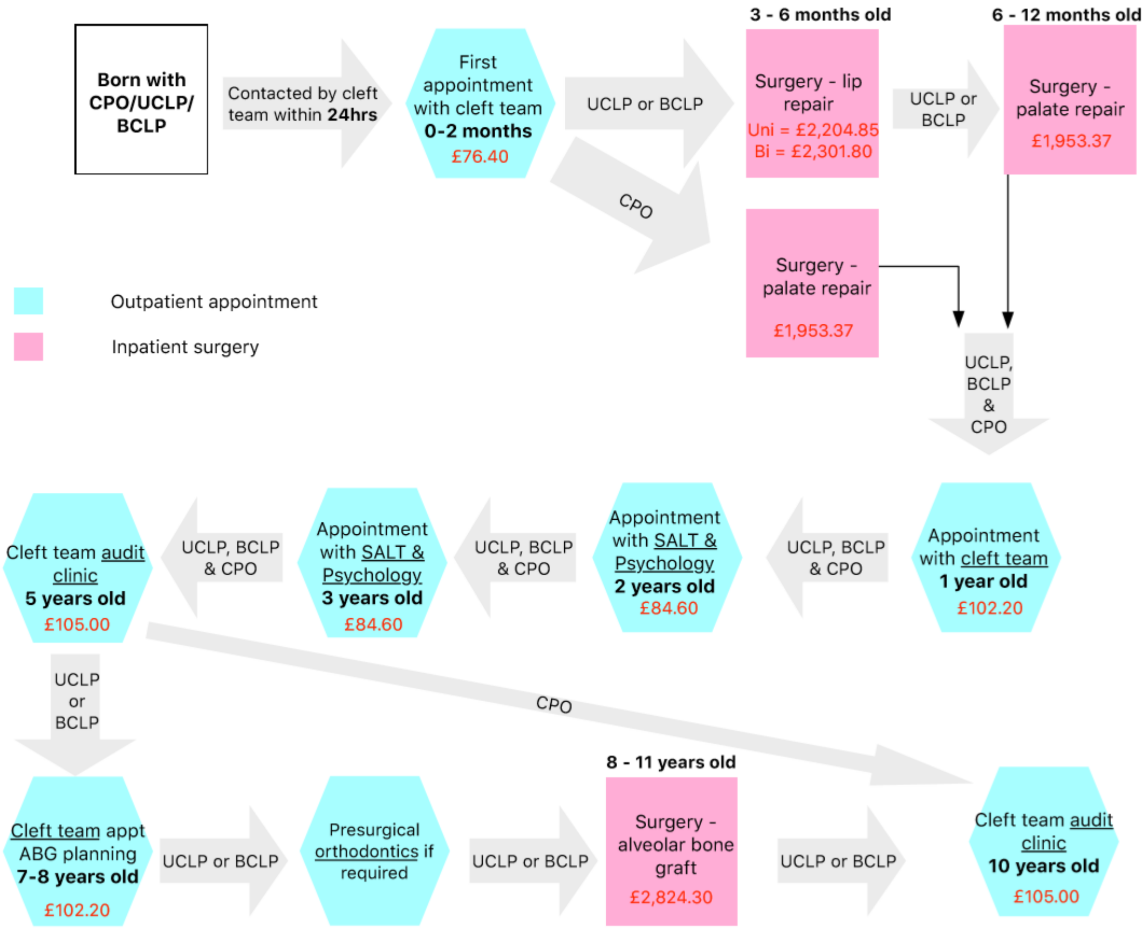

Figure 14. Simplified version of typical cleft care pathway of the 3 cleft phenotypes between ages of birth and 10 years old with mean calculated costings applied. Modified from South Wales South West Managed Clinical Network – Typical Patient Journey flowcharts (2016)

6.4 Comparisons to other studies

When costings from this study are compared to other studies, then there are differences, but it is important to note where these differences lie. A study which estimated national averages of revision surgeries for orofacial clefts in the United States of America (USA) has shown that the mean cost per hospitalisation ranged from $7564 (£6018) to $8393 (£6678) from the cost year of 2009 (Thompson et al., 2017) (Currency conversion at rate of 0.80 GBP to 1 USD on 19/07/2020). These costs when inflated using Bank of England inflation calculator were £7930 to £8799. A similar surgery cost found from the collected data would range from £1565.17 (palate re repair mean cost) to £1837.05 (lip re repair cost). These differences can be attributed to differences in staffing numbers and staff salaries in different countries. Funding is also different in other parts of the world, with the USA being a more insurance led model. This is an important point as the study goes on to note that there is a 1.71 odds ratio (95% CI: 1.41—2.09) of the patient having revision surgery of the lip if the patient has insurance. It was noted that results were similar for the palate too. This is higher than the odds ratio of a syndromic diagnosis requiring revision surgery of the lip (OR 1.47, 95% CI: 1.16-1.87). In the data found from our study, revision surgeries were not as common as described in this study from the USA, most probably as the NHS is a publicly funded healthcare system, where money is less likely to be a part of the decision whether or not to consider a surgical revision. Instead, any such decision is more clinically driven.

Kumar *et al.* (2006) carried out a study in the South West of England which micro costed orthognathic surgery and the components within the treatment pathway. This included joint orthodontic maxillofacial surgery outpatient clinics being costed at a total of 160 Euros (£146, inflated cost using Bank of England inflation calculator £237) (conversion rate on 19/07/2020 at £0.91/1 Euro). This is comparable to the joint cleft outpatient clinic where the cost was found to be £102.20 (£93.05 to £110.90 from the lowest to the highest salary points). The additional costs in the joint clinic in the orthognathic surgery study are attributed to having more trainees or additional consultants attending rather than SALT and psychology team members or geographical differences in hospital costs. The study for orthognathic surgery costed the total inpatient and surgery costs for the procedure at 3327.06 Euros (£3026, inflated cost using Bank of England inflation calculator £4916) (conversion rate on 19/07/2020 at £0.91/1 Euro) (Kumar *et al.,* 2006), which is comparable to the cost found for ABG surgery in the UCLP and BCLP groups studied here (£2,824.30). The study for orthognathic surgery also included provision of high dependency beds where required, which would have increased the costs significantly. High dependency and paediatric intensive care bed costs have not been factored in this cleft care study as the studied case notes showed that none of the patients required this level of care. If this was required, it would add to the surgical costs significantly. Interestingly, the study also noted that the surgical costs could vary between units, with a range of 5312.26 Euros to 7798.50 Euros (£4832 to £7093, inflated cost using Bank of England inflation calculator £7851 to £11524) (conversion rate on 19/07/2020 at £0.91/1 Euro) (Kumar *et al.,* 2006). This might suggest that the costs found here for cleft care surgery appear to be a reasonable representation of the patient costs incurred.

Complications during surgery are risks that apply to all surgical procedures. Surgical complications can increase operating time and may indicate the need for revision surgery to improve function, aesthetics and/or additional outpatient care provision. Revision surgery in this study had a mean cost of £1,837.05 and £1,565.17 for lip revision and palate re-repair respectively. Complications also result in an increased hospital length of stay in a study from the USA on cleft affected individuals (Nguyen *et al.,* 2014). From the data collected in the present study it was seen that for each extra hospital day of stay there is an added cost of £286.34 and so additional days can increase the costs significantly. These costs would be higher should this bed be required to have high dependency care provision. High dependency care or intensive care provision is a consideration in cleft affected individuals, but tends to be in cases where there may be potential airway management risk. These risks are present in some syndromic cases such as Pierre Robin (Oxford Handbook of Clinical Skills for Children's and Young People's Nursing, 2012).

Team familiarity can play a role in efficiency. If a team has a set way of working over a number of years, through audit, teaching and open discussions, efficiency can improve and may be evidenced by decreased surgical time. This has been shown in other medical surgical fields. In a study of laparoscopic surgery with 100 cases treated by either a familiar certified surgical assistant (CSA) or a new CSA in the USA the less experienced operator demonstrated much longer associated operative times. It also went on to state "despite confinement to a single surgeon and procedure, these results suggest what all surgeons know: excellent help is priceless" (Finnesgard *et al.,* 2018). This is important to note, as a replacement of any member of the team may risk increasing surgical times, and can explain why there can be a large range of times seen in these surgeries, and operating times may vary from operator to operator.

An issue with micro costing is that quality of care is not necessarily measured as there is no numerical cost to directly assign for this, nor can it be identified from the clinical notes. However, although revision surgery was common it may not necessarily be associated with poor quality treatment. Some revision surgery may take place when the individual's soft tissues have matured, or when patients reach puberty and aesthetics may be of more concern to them. Outcomes of surgery in this study were not assessed for aesthetics and function and so quality is a factor that was not assessed. Should cost-effectiveness be investigated, quality of care would need to be taken into account.

Overheads costs for outpatient clinics were assumed to include personal protective equipment (PPE) costs. This was a consumable cost included in the overheads for the clinics. These costs were nominal due to the low cost of these products and the large volumes in which they are purchased. To prevent over costing this was omitted from being additional to the clinical overheads. The latter were costed prior to the new guidelines which now exist because of COVID-19. These guidelines, which are ever changing, but at the time of writing included the need to use facemasks in all hospital areas with enhanced equipment required if there was an increased risk identified. Costs of masks, gloves and aprons have increased significantly (varying from suppliers), and in future studies these costs would need to carry a higher weighting to factor the increased costs. By way of illustration, suitable disposable masks (N95) now cost $1.50 (£1.20) each, which is a significant increase from the masks previously used which cost less than five pence each (Mick and Murphy*.,* 2020). Overhead costs for sterilisation of equipment for outpatient appointments were costed at £20, a cost provided by the central sterilisation services at the UHBW. This is a reasonable cost when compared to other departments, as found in University Hospitals Birmingham freedom of information requests document in 2015 (University Hospitals Birmingham, 2015).

The majority of cleft affected individuals have some psychological input in joint clinics, but rarely go on to need further treatment. The CRANE report of 2018 (CRANE, 2018) showed that only 10% of the cleft affected individuals assessed required more treatment with a psychologist other than initial consultations in joint cleft clinics. This corroborates with found data, as minimal psychological input was found from the clinical notes.

6.5 Data collection

Data was collected from 23 sets of case notes (5 BCLP; 8 CPO; 10 UCLP). The reason for this were insufficient records being present. Following the CSAG report, cleft treatment in the UK underwent major reorganisation in order to create a centralised service with the aim to improve patient outcomes. In order to ensure that the records identified were of children who had been through the new centralised system, patients were selected between 2015 and 2018 where case notes were available.

Studies involving data collection from hospital notes are known to struggle with data extraction. Burnett *et al.* (2011) found that in 15% of 1,161 cases studied, key outpatient clinical information was missing from consultations. The study was based in three London teaching hospitals and went on to discuss that having an electronic medical record would reduce the incidence of missing clinical information, and concluded that regular auditing would also reduce the prevalence of missing information. This is a problem in other countries too, as highlighted by a study in the USA where 13.6% of clinicians reported there was missing data from 1614 patient notes assessed. This included missing letters, laboratory results, history and examination notes and medications for the patient (Smith *et al.,* 2005).

Ideally, a digital centralised system would improve access to patient data. There is a national drive towards digital note keeping throughout the NHS, but previous attempts to do this has cost billions of pounds and failed (House of Commons Committee of Public Accounts, 2011). A centralised system would allow easier and more comprehensive access to case notes, as well as remote access to the data. For the purposes of this retrospective study, the data collection involved accessing and assessing:

- General clinical notes,
- SALT notes,
- ENT notes from St Michael's hospital,
- Digital notes checked where possible to cross reference with letters sent to ensure no clinics were missed.

In future, a digital note keeping system should allow a more robust data collection process. Another more robust way of collecting accurate data would be to carry this project out prospectively, using a customised data collection sheet with drop down lists in order to ease input and subsequent data analysis. Electronic data collection has been shown to be easier to execute and to analyse, but with these benefits come other caveats such as specific equipment required and security of devices (Dickinson *et al.,* 2019). Despite the limitations with the non-centralised and non-digital infrastructure, every effort was made to ensure that a comprehensive dataset was compiled. By cross referencing the data sets, all data that could have been found were extracted and assessed to ensure there was no duplication was present. This involved considerable time and effort to ensure the data were an accurate representation of information from the clinical notes.

6.5.1 Inpatient data collection

There was a wide range in the time taken for surgery, which affected the observed costs, and in order to better understand this, discussions were had with a cleft orthodontic consultant. The possible reasons for these variations were considered to be: operator variability, difficulty of anaesthesia, complications during the surgery and differences in case complexity.

Different operators may use different techniques, which may vary in the time taken to carry out the treatment, in turn adding a range of costs. Experience of the surgeon will also affect the time taken to carry out a procedure. This was highlighted by a study which reviewed a number of other medical specialty operations including coronary artery bypass grafting surgery, knee replacements and bilateral reduction mammoplasties in a tertiary care academic hospital in the USA. The study reported that there was a significant reduction in operating times associated with surgeon experience. It went on to show that after 15 years of experience, surgical times decreased by 8% to 48% depending on the specific procedure (Maruthappu *et al.,* 2015).

Anaesthetic medication is administered by the anaesthetist with the assistance of the anaesthetic nurse and often an anaesthetic trainee. Cases of cleft surgery may be a particularly important training experience, as it is likely that it may be one of a small number of such cases the trainee will see during their training. This may increase the time taken for surgeries on cleft affected individuals and this added time has its own associated costs.

Initially when data were collected, data were extracted from the notes. Every attempt was made to make the data as accurate as possible. Data were collected retrospectively, and in some cases not all surgical or inpatient data were available. Surgical data were predominantly collected from hospital ward records in the case notes and anaesthetic charts. The latter record the time that anaesthetic medication is given until the end of the operation. Recovery time is also noted on the anaesthetic chart. This does not cover the time taken to write up notes, to have surgical checklists carried out, nor does it include time taken to reflect on the case as clinicians often do. This ‘buffer time’ was deemed to be 20 minutes either side of the surgery when discussed with a number of consultant maxillofacial surgeons and oral surgeons. This is supported by a study in a teaching hospital in England which found that a median time of 22.5 minutes wait was found either side of 55 observed surgeries over 22 operating lists of gynaecology surgery (Saha *et al.,* 2009). With certain complex cases, or those where different equipment was used, this may have been an underestimate of the time and cost. However, as this could not be found retrospectively it was deemed best to be conservative with the estimation.

Where surgical/inpatient duration data were not available, average times were applied to these specific cases. Mean unilateral lip repair times were 127 minutes in BCLP, whilst UCLP mean unilateral lip repair times were 126 minutes. Owing to the similar times, a mean unilateral lip repair was applied. Differences between phenotypes were minimal, and due to limited numbers of BCLP cases, mean times were used. This was the mean substitution method for managing missing data (Kang, 2013), in order to utilise the data found and apply it to data that were not available. Mean times were calculated for surgeries of each classification using data found from all of the other studied cases of the same classification. This was deemed as a fair way of giving a mean surgery time for similar cohorts of patients with similar operators. No cases were admitted to a high dependency unit or intensive care unit. This may have been the case for other treated cases and perhaps having a larger sample would have brought these costs into the means. If the surgical stay duration was not available then an average hospital duration (derived from averaging other similar surgical stays) was added. This was one night for each of the surgeries, and did not vary between phenotypes.

Grommets operation sheets were not found in any of the case notes but were found to have been carried out from letters from ENT or audiology. Whenever it was noted that grommets were inserted, an assumed cost found from the literature (Evidence Note 22, 2008) was applied and was inflated to the costs that would be incurred at the present time using the Bank of England calculator for inflation (Bank of England, 2020). The cost estimated from literature for grommets was (pre inflated literature cost £853.20) £1192.42 as a day case. This was the lowest mean cost of surgery across all the surgeries that were micro costed. The figure seems reasonable based on costs of surgery found in our study, of which the next lowest cost of surgical input was palate re-repair costing £1565.17 (with a night in the ward included at £286.34).

Pharyngoplasty operation was only noted in one set of case notes. A retrospective analysis of 300 consecutive patients in Manitoba who presented from 1980–1995 found that 25% of cases had a subsequent pharyngoplasty following primary palate repair (Bicknell et al., 2002). The wider literature suggests that between 15-45% of cases had a pharyngoplasty and that the precise anatomical location of the cleft heavily affected the need for this operation (Marinnan et al., 1998 and Mackay et al., 1999). Only one pharyngoplasty was noted which is not representative of the literature but may be ascribed to surgical operative variation from country to country, or skilled SALT teams working with the patients to avoid the need for this surgery. Another more likely reason is that the ‘palate re repair’ surgeries may have had a pharyngoplasty carried out as part of the procedure. However, as these cases were not specifically labelled as a pharyngoplasty, from the data it appears to be an uncommon procedure.

One of the UCLP cases had their surgeries carried out in Wales. The data for time spent under anaesthetic and in recovery was not available. However, the number of days spent in hospital was available. Funding is different in NHS Wales when compared with NHS England, and can vary between Trusts. As a result, the number of days of hospital stay was costed using the studied hospital (Bristol Royal Children's Hospital) costs, applying average times found for similar surgeries in this study to provide a fair representation of the costs. A further case had treatment carried out in another hospital in England. This case had the Bristol Royal Children's Hospital costs applied to the surgical times and hospital stay duration which was a fair cost comparison.

Surgical consumables were costed using theatre preparation lists for operative procedures. These lists were constructed by the theatre staff and the operating consultants locally in order to ensure that equipment was ready for when surgery began. Attendance at the surgical procedures confirmed that these lists were a true reflection of the equipment provided for each of the surgeries observed. Some of the consumables may have a short shelf-life but still require an inventory to be held should they be required for procedures. The cost for outdated consumables or wastage was not factored in this micro costing analysis.

A list was obtained from the finance department providing details of overheads. This included clinical area costs and maintenance of these areas, insurances, equipment, training, servicing costs, general staffing and utilities. Initial outlay of expenses for equipment are taken into account in these calculations, however this is applicable to the hospital generally.

The mean overall costs for treatment of a cleft affected individual from birth to 10 years of age were found to be £17,00.09 (SD £7,361.83), £6,137.49 (SD £2,319.87) and £11,619.74 (SD £2,547.81) for BCLP, CPO and UCLP respectively. The mean cost of treatment for CPO was found to be lower than that of UCLP, which was lower than that of BCLP. This was an expected result, as the care pathway for CPO had less surgical input than both UCLP and BCLP. The surgical input for UCLP would be expected to be less than BCLP as the initial cleft may be less complex or managed in one operation as it is unilateral. Outpatient input for these cases can vary considerably and may include input from the community setting.

6.6 Health resource group (HRG) code comparisons

HRG codes are used for reimbursing the relevant departments associated with the activities that have been carried out. There are numerous procedure codes assigned for each different procedure or activity, which apply to both inpatient and outpatient activities. Due to the vast number of codes present for different procedures, the codes are grouped using a grouping tool which form HRG codes. These groupings describe several procedures or activities that are deemed to be similar in order to assign a reference cost for each of these groupings. The reference costs are based on national averages and are applicable for particular cost years in order to find costs for delivering each service. The amounts reimbursed vary based on whether the activity was inpatient, outpatient or a day case and is affected by age range and comorbidities. The costs found for the inpatient or surgical procedures carried out on individuals with orofacial clefting up until the age of 10 years old have been compared in Table 26 to the HRG costs of each procedure.

The HRG costs (NHS Digital – National Reference Costs 2017- 2018 and tariff costs from National tariff workbook 2017 - 2018) are higher than the costs found from this micro costing study (Table 23 and Table 24). The variability in costs nationally may be due to a number of factors including:

- Operator variation – some operators may be more efficient than others
- Technique differences
- Experience of operator
- Number of other scheduled cases on an operating list
- Number of staff present including number of trainees and their stage of training
- Length of hospital stay – each additional night increases the costs significantly
- Associated comorbidities
- Age of patient
- Training opportunities during the activity

Procedures that could be classified as procedure description	Procedure description	OPCS descriptions	Relative HRG Descriptions	HRG Code	Cost of Each (Reference cost)	Cost of Each (Tariff) (Multiplied by MFF 1.084069)	Mean cost of procedure (at median salary point)
'Unilateral lip repair', 'bilateral lip repair'	Lip repair	Primary closure of cleft lip	Very Major, Mouth or Throat Procedures, 1 year and under	CA82D	£5,765	£3,659 (£3,966.61)	Unilateral lip repair £2,204.85 Bilateral lip repair £2,301.80
'lip re repair'	Lip repair revision	Revision of primary closure of cleft lip	Major, Mouth or Throat Procedures, 18 years and under	CA83C	£3,330	£1,502 (£1,628.27)	Lip re repair £1,837.05
'lip re repair'	Adjustment to vermilion border of lip NEC	Adjustment to vermilion border of lip NEC	Minor, Mouth or Throat Procedures, 2 - 18 years	CA85B	£1,732	£721 (£781.61)	Lip re repair £1,837.05
'palate repair'	Palate repair	Primary repair of cleft palate	Very Major, Mouth or Throat Procedures, 1 year and under	CA82D	£5,765	£3,659 (£3,966.61)	Palate re repair £1,565.17
'palate re repair'	Palate re repair	Revision of repair of cleft palate	Very Major, Mouth or Throat Procedures, 2 - 18 years	CA82C	£5,683	£2,992 (£3,243.53)	Palate re repair £1,565.17
'palate re repair'	Other specified correction of deformity of palate	Other specified correction of deformity of palate	Very Major, Mouth or Throat Procedures, 2 - 18 years	CA82C	£5,683	£2,992 (£3,243.53)	Palate re repair £1,565.17
'palate re repair'	Unspecified correction of deformity of palate	Unspecified correction of deformity of palate	Very Major, Mouth or Throat Procedures, 2 - 18 years	CA82C	£5,683	£2,992 (£3,243.53)	Palate re repair £1,565.17

‘palate re repair’	Suture of palate	Suture of palate	Minor, Mouth or Throat Procedures, 2 - 18 years	CA85B	£1,732	£721 (£781.61)	Palate re repair £1,565.17
‘palate re repair’	Other specified other repair of palate	Other specified other repair of palate	Very Major, Mouth or Throat Procedures, 2 - 18 years	CA82C	£5,683	£2,992 (£3,243.53)	Palate re repair £1,565.17
‘palate re repair’, ‘lip and palate re repair’	Unspecified other repair of palate	Unspecified other repair of palate	Minor, Mouth or Throat Procedures, 2 - 18 years	CA85B	£1,732	£721 (£781.61)	Palate re repair £1,565.17
‘Pharyngoplasty’	Pharyngoplasty	Plastic repair of pharynx NEC	Complex, Mouth or Throat Procedures, 2 - 18 years	CA81C	£4,837	£3,554 (£3,852.78)	Pharyngoplasty £1,713.88
‘Pharyngoplasty’	Pharyngoplasty	Other specified repair of pharynx	Complex, Mouth or Throat Procedures, 2 - 18 years	CA81C	£ 4,837	£3,554 (£3,852.78)	Pharyngoplasty £1,713.88
‘Pharyngoplasty’	Pharyngoplasty	Unspecified repair of pharynx	Complex, Mouth or Throat Procedures, 2 - 18 years	CA81C	£4,837	£3,554 (£3,852.78)	Pharyngoplasty £1,713.88
‘ABG’	ABG	Augmentation of alveolar ridge NEC	Major dental procedures	CD01B	£3,892	£960 (£1,040.71)	ABG £2,824.30
‘ABG’	ABG	Alveolar bone graft to maxilla	Intermediate MF procedures	CA94	£4,137	£1,904 (£2,064.07)	ABG £2,824.30
‘ABG’	ABG	Augmentation of alveolar ridge using auto bone graft	Major Dental Procedures, 18 years and under	CD01B	£3,892	£960 (£1,040.71)	ABG £2,824.30

Table 23. HRG costs (NHS Digital – National Reference Costs 2017- 2018 and tariff costs from National tariff workbook 2017 - 2018) for the surgical/inpatient care compared to mean costs found from studied cases.

Outpatient Appointment	**cost of clinic @ median pay point/**	**Outpatient reference cost**	**Outpatient tariff cost (Multiplied by MFF 1.084069)**
Cleft clinic	£76.40	First attendance: £157; Follow up: £129	First attendance: £176 (£190.80); Follow up: £84 (£91.07)
Joint cleft clinic	£102.20	First attendance: £157; Follow up: £129	First attendance: £176 (£190.80); Follow up: £84 (£91.07)
Audit clinic	£105	First attendance: £157; Follow up: £129	First attendance: £176 (£190.80); Follow up: £84 (£91.07)
Speech investigation clinic	£78.20	First attendance: £122; Follow up: £120	First attendance: £149 (£161.53); Follow up: £77 (£83.47)
Cleft nurse review-in person	£43.10	First attendance: £88; Follow up: £97	No tariff cost available
Cleft nurse phone review	£15.40	First attendance: £137; Follow up: £127	No tariff cost available
Cleft nurse home visit	£92.40	First attendance: £88; Follow up: £97	No tariff cost available
Paediatric dentistry	£73	First attendance: £154; Follow up: £145	No tariff cost available
Audiology	£49.25	First attendance: £96; Follow up: £116	No tariff cost available
SALT (face to face)	£48.60	First attendance: £115; Follow up: £100	No tariff cost available
SALT telephone	£24.30	First attendance: £67; Follow up: £53	No tariff cost available
SALT group therapy	£24.30	First attendance: £115; Follow up: £100	No tariff cost available
SALT school visit	£97.20	First attendance: £115; Follow up: £100	No tariff cost available
SALT and Psychology	£84.60	First attendance: £103; Follow up: £313	No tariff cost available
Photography	£24.30	No reference cost available	No tariff cost available
Psychology	£63.30	First attendance: £315; Follow up: £298	No tariff cost available

Table 24. HRG costs (NHS Digital – National Reference Costs 2017- 2018 and tariff costs from National tariff workbook 2017 - 2018) for the outpatient secondary care compared to mean costs found from cases.

Reference costs are costs that are based on national averages of historical data and are not the values that are provided to the hospital in order to provide the service. Tariff costs are the amounts that are provided to the hospital for delivering the specified clinical activity after adjustments have been made due to reference costs being three years in arrears when produced. As a result, the reference costs, being national averages do not require market force factors (MFF) to be applied to the values. The MFF value assigned to University Hospital Bristol was 1.084069. This MFF value is a required adjustment to the tariff cost and is multiplied by tariff costs in order to compensate for the differences in costs that is found in different parts of the country and between the providers of healthcare. Tariff costs are also affected by best clinical practice as opposed to just average costings.

An important point to note is that the HRG codes are not costed differently for individuals who present with associated syndromes when below 18 years old. Patients who presented with a syndrome were excluded from this study as results may have not been as generalisable due to the large variation in syndromes in which clefting is a feature and the vast differences in management depending on the syndrome. This may account for the reason HRG reference costs were found to be higher than the those determined using micro-costing here, as they were based on an average from historical data of patients. Tariff costs have been found to be closer to the calculated costs from our study.

6.7 Changes to practice

This study has shown the mean costs for the provision of care of three cleft phenotypes. The costs found for each of the surgical procedures or outpatient activities could be used as a benchmark of costs when commissioning the cleft service. The study also highlights that surgical costs make up a larger proportion of the costs than outpatient activity. The results from this study together with a parallel study by Durman (2020) investigating similar costs for the same phenotypes but between 10-20 years, could be used to develop an evidence based protected budget for cleft care provision.

Another recommendation for practice would be the development of a centralised clinical notes system. This would be useful for audit trails, and to minimise missing data. It would also mean that data could be accessed remotely once the infrastructure is in place. The NHS is moving to a minimal paper system, which will not only improve research and audit processes but will also benefit the environment.

7 Conclusion

This micro costing analysis has found that the mean (SD) costs from birth to 10 years of age for provision of cleft care for each phenotype were:

- BCLP - £17,004.09 (SD £7,361.83)
- CPO - £6,137.49 (SD £2,319.87)
- UCLP - £11,619.74 (SD £2,547.81)

The main costs were derived from inpatient and surgical care of patients. Revision surgeries cost less than the initial surgical procedure, but these were still substantial. An example of this was unilateral lip re repair (£1837.05) being only £367.80 less than the initial surgical intervention (£2204.85). The surgical procedure with the highest cost was alveolar bone graft surgery costing £2,824.30.

Breakdowns of the micro costing found from this study showed that staff costs formed the bulk of the costs of both outpatient appointments and in surgical or inpatient care. Pharmaceutical costs were the lowest costs to the care pathway. Outpatient activity made up the largest proportion activity, with SALT having the most frequent patient contact.

When compared with the National Tariffs, costs were lower and this likely to be in part attributable to the lack of a mechanism for coding patients with comorbidities under the age of 18 years.

Although the findings of this study may be applied to other centres, costs can vary significantly between different Trusts and even more so when comparing with other countries.

7.1 Future studies

Previous work carried out by Souster (2017) identified the need to micro cost the provision of care to 10 years old and not just in UCLP but also BCLP and CPO. The current project paralleled the study by Durman (2020), who micro-costed the provision of care between 10 and 20 years of age for the same three phenotypes. Together, these projects will provide a micro costing of the provision of care from birth to 20 years old for the three cleft phenotypes.

With the advent of electronic patient records it may be easier for future studies to be run prospectively, continuously and in a less labour-intensive way. This will mean that any micro costing can remain contemporary and easily accessible almost in real time.

Micro costing should be considered for the treatment of the most common syndromic cases to provide a more embracing average cost for cleft care. This would also bring costs closer to the HRG tariffs.

An interesting follow up study that could be carried out would be to evaluate HRG coded costs for a patient with orofacial clefting from birth to 10 years of age and compare this with micro costed values. Differences between the two could be further investigated in order to develop more accurate codes or costings. The limitations of the study might also be mitigated, such as seeking ethical approval could be sought to examine community aspects of care as well as the prenatal aspects of the provision of cleft care to the whole family.

Carrying this study out in other units, or even using costs from finance departments of other hospitals could provide a range of costs in which the national costs could be developed for the provision of cleft care. If this were extended to Wales and Scotland, it could help with budget allocation in these countries too.

Table 25 highlights both the known and the unknown costs found in this micro costing study. A further study might identify more of the unknown costs to provide more accurate costings for the provision of cleft care for the specified ages.

Known Costs	**Unknown Costs**
Birth associated costs e.g. cleft nurse review	Pre-natal costs
Outpatient MDT clinic costs	Community audiology, psychology, dental
Outpatient single specialty clinic costs	ENT outpatient data - some notes missing
Outpatient SALT appointment costs	Precise day case grommet costs
Outpatient audit clinic costs	Staff travel costs
Hospital dental review costs	Special tests – radiographs, photography
Inpatient surgery costs	

Table 25. Known and unknown costs identified from this study.

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9. Appendices

Appendix I: Surgical consumables used in pharyngoplasty

Item	Price (£)	Number	Total (£)
cleft set	23.12	1	23.12
15 blade	0.05	6	0.3
hypos	0.01	3	0.03
5ml syringe	0.01	1	0.01
suction tubing	0.37	1	0.37
small raytec swabs	2.7	5	13.5
1.25cm throat pack	0.42	1	0.42
no 4 size patties	0.54	1	0.54
black suction catheter	0.05	2	0.1
light handles	0.05	2	0.1
ent drape pack	3.66	1	3.66
split sheet	3.08	1	3.08
bowl pack	1.98	1	1.98
4/0 vicryl W9067	2.06	1	2.06
2/0 silk W578	0.07	1	0.07
gloves	1.4	7	9.8
gown	1.7	7	11.9
mask with eye protection	2.3	7	16.1
antiseptic betadene	5.61	1	5.61
crystal violet ink	0.3	1	0.3

lignospan	0.39	2	0.78
bipolar lead	46	1	46
steristrips 1/2 "	0.03	1	0.03
yellow paraffin	0.59	1	0.59
hydrofilm	0.08	1	0.08
saline	3.13	2	6.26
sharps box	0.04	2	0.08
table cover	1.13	2	2.26
bipolar	30	2	60
dental syringe	0.01	1	0.01
Total cost (£)			**209.14**

Appendix II: Surgical consumables used in alveolar bone graft

Item	Price (£)	Number	Total (£)
oral tray	23.18	1	23.18
alveolar tray	6.47	1	6.47
cleft set	23.12	1	23.12
orthopaedic tray	22.21	1	22.21
bone nibbler	1	1	1
osteotomy tray	18.58	1	18.58
west retractor hip	1	1	1
T-bar handle & jacobs chuck	4.77	1	4.77
bowl pack	1.98	1	1.98
light handles	0.05	2	0.1
small raytec swabs	2.7	5	13.5
yanker sucker	0.05	1	0.05
frasier no9 sucker	2.7	2	5.4
sharps box	0.04	2	0.08
biogide	97.2	1	97.2
fine marker pen	0.03	2	0.06
4/0 vicryl W9067	2.06	1	2.06
15 blade	0.05	6	0.3
ent drape pack	3.66	2	7.32
split sheet	3.08	2	6.16
Dental syringe	0.01	2	0.02
5ml syringe	0.01	1	0.01

10ml leur lock syringe	0.01	2	0.02
hypos	0.01	3	0.03
bone wax	1.55	1	1.55
3/0 vicryl rapide	2.2	2	4.4
4/0 vicryl W9067	2.06	1	2.06
Antiseptic betadene	5.61	1	5.61
bipolar	30	2	60
bipolar lead	46	2	92
saline	3.13	2	6.26
Antiseptic betadene	5.61	1	5.61
Yellow paraffin	0.59	1	0.59
lignospan	0.39	3	1.17
gown	1.7	7	11.9
mask with eye protection	2.3	7	16.1
suction tubing	0.37	1	0.37
1.25cm throat pack	0.42	1	0.42
Steristrips 1/2 "	0.03	1	0.03
monopolar	1.87	1	1.87
table cover	1.13	2	2.26
DBX putty	82.77	1	82.77
grafton	282	1	282
0.5mm permcol	288	1	288
gloves	1.4	7	9.8
Total cost (£)			**1099.59**

Appendix III: Surgical consumables used in cleft palate repair

Item	Price (£)	Number	Total (£)
cleft set	23	1	23
light handles	0	2	0.1
ent drape pack	4	1	3.7
15 blade	0	6	0.3
5ml syringe	0	1	0
10ml leur lock syringe	0	2	0
retrobulbar needle	0	1	0
filter needles	0	2	0
small raytec swabs	3	5	14
sharps box	0	1	0
gown	2	7	12
mask with eye protection	2	7	16
suction tubing	0	1	0.4
1.25cm throat pack	0	1	0.4
steristrips 1/2 "	0	1	0
nasal swabs	1	4	5
microscope handle covers	4	1	3.6
micro pattie (1/4" x 1/4" yellow string)	1	1	1.1
60 degree beaver blade	8	1	7.7
4/0 vicryl W9067	2	1	2.1

4/0 PDS round	2	1	2.2
5/0 pds	2	1	2.2
5/0 vicryl round	2	1	1.7
4/0 prolene	1	1	1.3
antiseptic betadene	6	1	5.6
bipolar	30	2	60
bipolar lead	46	2	92
saline	3	2	6.3
antiseptic betadene	6	1	5.6
Yellow paraffin	1	1	0.6
lignospan	0	3	1.2
crystal violet ink	0	1	0.3
monopolar	2	1	1.9
yanker sucker	0	1	0.1
fine marker pen	0	1	0
hypos	0	3	0
dental syringe	0	2	0
table cover	1	2	2.3
gloves	1	7	9.8
Total cost (£)			**282**

Appendix IV: Surgical consumables used in cleft lip repair

Item	Price (£)	Number	Total (£)
cleft set	23.12	1	23.12
light handles	0.05	2	0.1
suction tubing	0.37	1	0.37
1.25cm throat pack	0.42	1	0.42
small raytec swabs	2.7	5	13.5
sharps box	0.04	1	0.04
filter needles	0.02	2	0.04
5ml syringe	0.01	1	0.01
ophthalmic blade	4.05	1	4.05
nasal swabs	1.25	8	10
60 degree beaver blade	7.67	1	7.67
ent drape pack	3.66	1	3.66
antiseptic betadene	5.61	1	5.61
bipolar	30	2	60
bipolar lead	46	2	92
saline	3.13	2	6.26
antiseptic betadene	5.61	1	5.61
yellow paraffin	0.59	1	0.59
lignospan	0.39	2	0.78
yankeur sucker	0.05	1	0.05
fine marker pen	0.03	1	0.03
steristrips 1/2 "	0.03	1	0.03

gown	1.7	7	11.9
mask with eye protection	2.3	7	16.1
hypos	0.01	3	0.03
dental syringe	0.01	1	0.01
monopolar	1.87	1	1.87
6/0 monocryl reverse cut	2.2	1	2.2
7/0 vicryle rapide	2.2	1	2.2
dermabond	17	1	17
5/0 vicryl round	1.7	1	1.7
4/0 PDS round	2.19	1	2.19
saline	3.13	2	6.26
bowl pack	1.98	1	1.98
blue suction catheter no. 8	0.05	1	0.05
table cover	1.13	2	2.26
15 blade	0.05	6	0.3
gloves	1.4	7	9.8
Total cost (£)			**309.79**

Appendix V: Anaesthetic medications and costs associated used in the surgical procedures

Anaesthetic medication	Cost (£)
Fentanyl (500mg)	0.80
Propofol (200mg)	1.50
Atracurium (50mg)	2.20
Ondansetron (4mg)	0.12
Dexamethasone (6.6mg)	2.00
Co-amoxiclav (1.2g)	0.90
IV paracetamol (1000mg)	1.20
Hartmann’s (1 litre)	0.91
Sevoflurane (/hour)	5.00

Appendix IV: ‘To take away’ (TTA) medications and costs

TTA's	Cost (£)
Paracetamol (32)	0.73
Ibuprofen (24)	0.66
Chlorhexidine (300 ml bottle)	4.09
Co-amoxiclav 500/125 (21)	1.84
Total	**7.32**